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Significant Conversations with Parents:

A systematic review of interventions to support the communication of bad news in paediatric settings and a qualitative study of parental experiences of receiving a newborn diagnosis of cystic fibrosis.

Bláthnaid Greene

Doctorate in Clinical Psychology
The University of Edinburgh
March 2020

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Thesis Portfolio Abstract

Background: Significant conversations with parents are a necessary and important part of healthcare. The delivery of bad or difficult news, such as the diagnosis of a chronic health condition, requires key communication skills. This is especially true for communication between healthcare professionals and parents in paediatric settings. It is important to understand parental experiences of these conversations and what can be done to help support and improve skills in this area.

Method: A systematic review evaluates studies assessing the effectiveness of interventions in improving breaking bad news skills. An empirical study uses Interpretative Phenomenological Analysis of interviews with parents to gain an in-depth understanding of parental experiences of receiving a newborn diagnosis of cystic fibrosis (CF).

Results: The systematic review identified ten quantitative studies assessed to be of either moderate or high quality. Significant improvements in communication skills were found following nine of the ten interventions. These interventions shared some common features. In the empirical study, three superordinate themes emerged following interview analysis: Cognitive and Emotional Experiences; Connection; and Knowledge.

Conclusions: Findings from the review suggest that there are interventions that can improve communication skills in delivering bad news in paediatric settings. Parents in the empirical study clearly recalled the period of receiving a newborn diagnosis of CF as an emotional time. Health professionals' communication and interpersonal skills seemed to play an important role in providing containment for families. Clinical implications and directions for future research are discussed.

Thesis Portfolio Lay Summary

Professionals working in healthcare need to deliver a range of important information to families, often requiring significant conversations. Breaking bad or difficult news, such as informing someone of a death or telling a parent that their child has a long-term illness, is a necessary part of healthcare, but having these kinds of conversations can be difficult. This is especially true where children and families are involved. Healthcare professionals have reported finding it stressful and not feeling confident when breaking bad news. Some research has found that parents may have different opinions to healthcare professionals when it comes to how bad news should be delivered. For this reason, and because parents are often the ones receiving this kind of news, it is important to hear about their experiences as well.

The first part of this thesis reviewed and evaluated scientific research looking at ways to improve communication skills when breaking bad news to parents. In nine of the ten research studies reviewed, interventions improved communication skills in healthcare professionals, suggesting that the right training can in fact improve these skills. Specific activities, such as using role plays or teaching skills while providing sufficient background information, were common features in these interventions. These should be considered in future training programmes for healthcare professionals.

The second part of this thesis is a study examining how parents experienced their child being diagnosed with cystic fibrosis after newborn screening. Interviews with seven mothers were analysed, and three common themes emerged: 1) diagnosis is

an emotional time for parents and is an event they remember clearly; 2) support and empathy are important to parents and they value connection with others. This can be with people they are already close to as well as new relationships with healthcare professionals; 3) there is a lot that parents need to learn and process following diagnosis. The type of information parents receive and the way this is shared with them is important. There may not be one approach that suits everyone and professionals need to be flexible to meet the needs of families. The final part of this study looks at how these themes link with other research.

Overall, communication and people skills are important in these kinds of significant conversations. In order to help and support families when they are receiving bad news, healthcare professionals need to think carefully about how they communicate information.

Interventions to support the communication of bad news in paediatric settings – a systematic review

Bláthnaid Greene^{1,2,*}, Mark Hoelterhoff², Shona Murphy¹, Kara Gibson³

¹Psychology Department, NHS Fife, Dunfermline, UK

²School of Health in Social Science, University of Edinburgh, Edinburgh, UK

³Child and Adolescent Mental Health Services, NHS Lanarkshire, Hamilton, UK

*Corresponding author: Bláthnaid Greene, contact email: blathnaidgreene@nhs.net

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Abstract

Background: Breaking bad news is a necessary part of healthcare. It requires key communication skills and has been identified as an area in which healthcare professionals may lack confidence, especially in paediatric settings. This study aims to systematically review the evidence of interventions to support and improve the communication of bad news by healthcare professionals in paediatric and children's services.

Method: A comprehensive literature search was conducted using three database platforms and a narrative synthesis of the findings is presented. Studies included used an observer rating of communication skills.

Results: Ten quantitative studies were identified as meeting inclusion criteria. Studies were assessed to be of either moderate or high quality. Significant improvements in communication skills were found following nine of the ten interventions. These interventions shared some common features.

Conclusions: Findings from this review suggest that training and intervention programmes can improve healthcare professionals' communication skills in delivering bad news in paediatric settings. Future interventions should consider including: role plays, didactic teaching and reflective practice. Directions for future research are also discussed.

Keywords: *Communication, bad news, paediatrics, interventions*

Introduction

The role of communication

Communication plays an essential and important role across all areas of healthcare. It has been identified as a key competency for physicians (e.g King & Hoppe, 2013) and is highlighted as a key skill in a range of guidelines (e.g. General Medical Council, 2013; National Institute for Health and Clinical Excellence, 2012). Effective communication between patients and clinicians has been linked to better health outcomes (Stewart, Meredith, Brown, & Galajda, 2000; Street Jr, Makoul, Arora, & Epstein, 2009) and greater patient satisfaction (Shaw, Zaia, Pransky, Winters, & Patterson, 2005; Wanzer, Booth-Butterfield, & Gruber, 2004). Communication skills have been identified as important for effective practice by doctors and as an area that can be improved through training (Choudhary & Gupta, 2015). A systematic review of the literature recognised that most qualified physicians receive substantial teaching on communication skills at various stages of training and beyond, and sought to identify strategies that were effective in supporting the development of communication skills (Berkhof, van Rijssen, Schellart, Anema, & van der Beek, 2011). From the studies examined, the authors concluded that active, practice-orientated strategies such as role plays, feedback and discussions were most beneficial.

Breaking bad news

Breaking “bad news” is one form of communication clinicians are often required to exercise (Johnson & Panagioti, 2018). Bad news can be defined as “any information likely to alter drastically a patient's view of his or her future” (Buckman, 1984, p.

1597). A range of vocabulary has been used to describe such information, including, but not limited to, “bad”, “negative” and “difficult” news. Furthermore, Ptacek and Eberhardt (1996) suggest that the extent to which news is viewed as bad may be linked to the negative emotional, psychological, and cognitive impact it has on the recipient, persisting over a duration of time following its delivery. The subjectivity associated with bad news has been highlighted with a recognition that it may be appraised as “bad” from multiple perspectives including the recipient, the news deliverer, or both (Fallowfield & Jenkins, 2004; Ptacek & Eberhardt, 1996). Whilst certain circumstances or events may be more objectively viewed as “bad news” by society (e.g. losing a child), a range of personal and other factors may influence how an individual interprets certain types of news.

Research to date has examined recommendations for delivering bad news, interventions to support communicating bad news, experiences of recipients (often patients or family members), and experiences of healthcare professionals delivering bad news (Ptacek & McIntosh, 2009). The considerable attention given to this area perhaps reflects the fact that, though unpleasant, delivering bad news is a necessary part of healthcare which needs to be supported by and monitored with effective evidence-based approaches (Fallowfield & Jenkins, 2004).

Healthcare professionals’ experiences of breaking bad news

Whilst breaking bad news can have psychosocial impacts on patients and families (Fallowfield & Jenkins, 2004), it can also affect the healthcare professionals delivering it. Breaking such news is recognised as a daunting experience and may

often be required before patients and healthcare professionals have developed a relationship (Monden, Gentry, & Cox, 2016). A lack of confidence in this area of communication has been self-reported by healthcare professionals (Dosanjh, Barnes, & Bhandari, 2001; Monden et al., 2016; Rider, Volkan, & Hafler, 2008). In a questionnaire-based study of paediatric residents, effective communication with patients was identified as a personal priority by 99% of respondents, though only 23% reported feeling confident in speaking to children about serious illness and only 27% reported feeling confident about giving bad news about a patient's illness to a patient and their family (Rider et al., 2008).

Some of the barriers to breaking bad news identified by residents include: a lack of emotional support and time, and clinicians' fears regarding their own abilities (Dosanjh et al., 2001). Increased levels of self-reported stress as well as physiological (cardiovascular) responses by medical staff and students are associated with breaking bad news in simulation settings (Brown et al., 2009; Hulsman et al., 2010; Shaw, Brown, Heinrich, & Dunn, 2013) and this is more likely for those who are fatigued or inexperienced in this skill (Brown et al., 2009). In a study looking at the trajectory of doctors' stress responses throughout a breaking bad news scenario, most participants showed a peak in heart rate response in the anticipatory phase of delivering the news with one third exhibiting a sustained response which did not return to baseline by the end of the simulated consultation (Shaw et al., 2013). Findings of these studies collecting self-reported and physiologically-measured responses emphasise the importance of considering the impact of breaking bad news on doctors. These effects can have longer-term consequences as well, and the relationships between stress, communication of bad

news and possible burnout warrant particular attention with research highlighting the prevalence of burnout and psychiatric comorbidity in doctors (Imo, 2017).

Breaking bad news in paediatric and children's services

Though there may be overlap in the skills and methods of breaking bad news across the lifespan and within different populations, work with children and families can bring its own specific considerations and challenges. In a study investigating medical residents' self-assessment of communication skills preparedness, residents reported receiving less training and feeling less prepared for paediatric compared to adult settings (Dubé, LaMonica, Boyle, Fuller, & Burkholder, 2003). This was the case for communication skills in general and specifically for those needed to deliver bad news.

Communication in paediatrics can be thought of as a triad between clinicians, parents, and their child, which may be complex and require skilled communication that is both patient- and family-centred (Levetown, 2008; Rider et al., 2008). Outside of this triad, other family members may also need to be considered (e.g. siblings, grandparents), as well as the wider systems within which children operate (e.g. school and peers). Investigating parents' appraisals of their child's healthcare, a study consisting of telephone interviews with 151 parents found that their perceptions of three factors were influential: clinicians' informativeness, interpersonal sensitivity, and partnership-building (Street, 1991). Levetown (2008) proposes two main areas of patient need during consultations: cognitive (relating to knowledge and understanding) and affective (involving emotional needs). Taking these concepts

together, Levetown states that physicians' behaviours must be both task-related and relational.

Studies in the fields of paediatric oncology and palliative care have highlighted the important role communication plays in parents' perception of high-quality care. Themes of parents valuing communication that is clear, honest, and demonstrates empathy and connectedness through relationship-building are echoed in studies examining the experiences and preferences of parents whose child has died (Davies & Connaughty, 2002; James & Johnson, 1997; Meyer, Ritholz, Burns, & Truog, 2006). In a study by Mack et al. (2005), providing clear information regarding end-of-life, communicating empathically with care and sensitivity, and communicating directly with their child were all factors significantly associated with higher parent ratings of care. The same study also examined physician-reported factors associated with high-quality care and found physicians' ratings of biomedical issues (such as child's pain and length of hospital stay) were significantly associated with higher care ratings whilst relational aspects were not.

Research gathering the perspectives of lay people and health professionals on breaking bad news directly to children asked participants to rate 64 story cards by how appropriate they felt the story was (Muñoz Sastre, Sorum, & Mullet, 2014). Lay perspectives were found to be more spread on a spectrum of what was viewed as appropriate (with some favouring saying little or nothing to the child), whereas health professionals' perspectives tended to cluster more around telling the child the truth. These findings emphasise the complexity of this issue and perhaps the important

role of practitioners' attunement to individual families. Whilst it is understandable that physicians may be more focused on medical and factual aspects, it is clear that the additional emotional needs and values of families must be considered as well.

Being attuned to the child, and their developmental and emotional stage is important when breaking bad news (Nunn, 2019). Nunn (2019) proposes that clinicians will need to take into account children's sensorimotor needs, receptiveness to action over words, and their use of play as language in the earlier years. As children become older, their growing number of attachment figures (through school and peers) will need to be held in mind, along with the possibility that they will increasingly seek comfort through friendships and more autonomous activities. They may, of course, move between these phases given what else is happening in their lives. Alongside this is the need to communicate effectively with parents who may be looking to the future and coming to terms with the contrast between their previous expectations and their current reality (Nunn, 2019). Just as no two patients would react the exact same way to the same news (Fallowfield & Jenkins, 2004), individuality exists within the family unit and each member may have a unique response (Nunn, 2019). This further emphasises the complexity of breaking bad news to families as health professionals may be required not only to tailor their approach between families but also within families, adapting to individuals.

Interventions to improve breaking of bad news

Whilst, historically, medical training may have placed more emphasis on "technical proficiency" (Monden et al., 2016, p. 101), growing emphasis appears to be placed

on communication in the field of healthcare. To support this, and the delivery of bad news in particular, a range of interventions aimed at practitioners have been described in the literature. Some of these include specific structured methods such as: the ABCDE technique (Rabow & Mcphee, 1999); the SPIKES protocol (Baile et al., 2000), and the BREAKS protocol (Narayanan, Bista, & Koshy, 2010). Each of these methods provides a mnemonic to assist practitioners in the delivery of bad news. ABCDE: *Advance* preparation, *Build* a therapeutic environment/relationship, *Communicate* well, *Deal* with patient and family reactions, and *Encourage* and validate emotions. SPIKES: *Setting* up the interview, assessing the patient's *Perception*, obtaining the patient's *Invitation*, giving *Knowledge* and information to the patient, addressing the patient's emotions with *Empathic* responses, and *Strategy and Summary*. BREAKS: *Background*, *Rapport*, *Exploring*, *Announce*, *Kindling*, and *Summarise*.

Fallowfield and Jenkins (2004) recognise that many seemingly appropriate recommendations and protocols exist, but research into their effectiveness has often relied upon self-report confidence ratings. They note that it is, however, important to understand whether interventions actually have a significant impact on clinicians' *behaviour*. One systematic review and meta-analysis of seventeen studies focused specifically on the effectiveness of interventions to improve breaking bad or difficult news across clinical healthcare settings (Johnson & Panagioti, 2018). Interventions were found to be significantly associated with large improvements in practitioners' breaking bad news skills and interventions based on the SPIKES protocol were found to bring about greater improvements than interventions based on other or no frameworks.

Rationale for this review:

Communication, particularly when it comes to delivering bad news, has been identified as an important area of consideration for practitioners, with additional challenges faced by those in paediatric settings. This review aims to build on previous research looking at interventions to improve communication of bad news, with a specific focus on approaches employed in the area of paediatric and child healthcare. It will address the question: “Which interventions improve professionals’ communication of bad news in paediatric settings?”. Communication of bad news can be with any individual linked to child (for example, the child or their parent/carer). In order to examine more objective communication measures, and acknowledge the limitations of self-report outcomes, the review will focus on studies which used observer ratings as outcome measures for communication. It is hoped this will contribute to the growing evidence base of effective approaches to support the communication of bad news in paediatric settings.

Methods

Search Strategy

Search terms were discussed with a librarian very familiar with searching the literature systematically and guided by a previous review looking at interventions across healthcare populations (Johnson & Panagioti, 2018). A protocol was written prior to formal searching (Appendix B). Searches were run through three database platforms: Ovid (PsychINFO, Embase classic+Embase, MEDLINE® Epub Ahead of Print, In-Process & Other Non-Indexed Citations and Daily), EBSCO (CINAHL and ERIC), and ProQuest (ASSIA, Dissertations and Theses Global) on 27th December 2019.

Below is the search strategy used in Ovid. A very similar search was conducted in the other database platforms with minor formatting changes where necessary.

1. "bad news"
2. (difficult adj (conversation* or news))
3. (negative adj (conversation* or news))
4. 1 or 2 or 3
5. (deliver* or inform* or communicat*)
6. 4 and 5
7. (paediatric* or pediatric* or child*)
8. 6 and 7

Eligibility Criteria

The Cochrane PICO search approach (Lefebvre et al., 2019) was used to guide the identification of studies that met inclusion criteria. Those which met the criteria for each of the areas listed below were included in this systematic review:

- **Population:** studies which included qualified or training clinical staff working in paediatric or children's services. Any settings related to paediatric or child services were included (e.g. children's hospital, paediatric emergency medicine).
- **Intervention:** studies examining interventions aimed at improving clinicians' communication of bad news to any person linked to the child (such as the child and/or their parent/carer). Any intervention formats would be reviewed including educational, skills training, specific protocols, and simulation.
- **Comparison:** studies with and without a control group. Those without a control group needed to at least include pre/post measures.
- **Outcomes:** studies which measured participants ability or skill in communicating bad news as rated by an observer. Any form of observer is included, for example, staff members, patients or simulated patients. Studies which also presented self-report findings are included but needed an observer rating as well.

Studies with quantitative intervention designs were included in this review. These could comprise any identified randomised control trials, non-randomised control trials, controlled before and after studies and interrupted time series as identified by

the Cochrane handbook (Higgins & Green, 2011). Less robust designs were also included for interventions not using designs listed above, though pre- and post-measures were required to be part of these studies. Google search engine was used to search for grey literature which may have met inclusion criteria for the review, no additional studies were identified using this approach. Only publications written in English were included.

Data Extraction

The first author (B.G.) extracted the data from publications that met inclusion criteria into an Excel (Microsoft) spreadsheet. Data extracted were split in to three overall categories: study characteristics, intervention descriptions, and outcome measures and findings. Study characteristics include: participants, sample size, service setting, country the intervention took place in, and research design. Intervention descriptions include: intervention used, theoretical basis for intervention, and use of 'expert' outside of research team (e.g. in healthcare area or expert by experience such as parent). Outcome measures and findings include: outcome measure(s) used, measurement time points, analysis and results, effect sizes, and findings summary. Effect sizes were calculated using Cohen's d. Where possible this was done manually using Cohen's d formula. Where data could not be sourced to use this formula, the "Practical Meta-Analysis Effect Size Calculator" (Wilson, 2020) was used and indicated in the results ((^ see Table 6).

Quality Assessment

Whilst considering quality assessment tools which could be applicable to the studies examined in this review, the authors of this paper reviewed the Downs and Black Checklist (Downs & Black, 1998). Recognising that there are areas of healthcare with few randomised controlled trials (the types traditionally reviewed in systematic reviews), Downs and Black developed this tool with a view to assessing both randomised and non-randomised studies.

The original Downs and Black checklist consists of 27 items with a maximum score of 32, enabling an overall quality score to be calculated for each study as well as a profile examining the quality in five areas: reporting, external validity, internal validity (bias), internal validity (confounding), and power. For all but two items, raters are required to make a decision between “yes”, “no”, and “unable to determine”, allocating a score of 1, 0 or 0 respectively. For the other two items they are required to provide a score of 0-2 or 0-5. The quality index is reported to have high internal consistency (Kuder-Richardson-20 = 0.89) and good test-retest reliability ($r=0.88$) and inter-rater reliability ($r=0.75$) (Downs & Black, 1998).

This checklist has been recognised as appropriate for those wishing to appraise research “quality and applicability to public health” (National Collaborating Centre for Methods and Tools, 2008, p. 1) and suitable for systematic reviews (Deeks et al., 2003). As the aim of this systematic review is to assess the methodological strength of these studies as opposed to assessing the quality of reporting in the papers, scores for the reporting area were excluded from final overall score calculations and

only those for internal validity, external validity, and power were included. As the Downs and Black Checklist does not provide score ranges corresponding to quality categories, the banding applied in a systematic review by Ratcliffe and colleagues (2013) was used. Based on the overall percentage score, studies were categorised as: low quality ($\leq 33.3\%$), moderate quality (33.4-66.7%) or high quality ($\geq 66.8\%$).

Table 1: Summary of quality assessment by area and criteria description

Area		Description
REPORTING	1	Is the hypothesis/aim/ objective of the study clearly described?
	2	Are the main outcomes to be measured clearly described in the Introduction or Methods section?
	3	Are the characteristics of the patients included in the study clearly described ?
	4	Are the interventions of interest clearly described?
	5	Are the distributions of principal confounders in each group of subjects to be compared clearly described?
	6	Are the main findings of the study clearly described?
	7	Does the study provide estimates of the random variability in the data for the main outcomes?
	8	Have all important adverse events that may be a consequence of the intervention been reported?
	9	Have the characteristics of patients lost to follow-up been described?
	10	Have actual probability values been reported (e.g. 0.035 rather than <0.05) for the main outcomes except where the probability value is less than 0.001?
EXTERNAL VALIDITY	11	Were the subjects asked to participate in the study representative of the entire population from which they were recruited?
	12	Were those subjects who were prepared to participate representative of the entire population from which they were recruited?
	13	Were the staff, places, and facilities where the patients were treated, representative of the treatment the majority of patients receive?
INTERNAL VALIDITY - BIAS	14	Was an attempt made to blind study subjects to the intervention they have received ?
	15	Was an attempt made to blind those measuring the main outcomes of the intervention?
	16	If any of the results of the study were based on "data dredging", was this made clear?
	17	In trials and cohort studies, do the analyses adjust for different lengths of follow-up of patients, or in case-control studies, is the time period between the intervention and outcome the same for cases and controls ?
	18	Were the statistical tests used to assess the main outcomes appropriate?
	19	Was compliance with the intervention/s reliable?
	20	Were the main outcome measures used accurate (valid and reliable)?
INTERNAL VALIDITY - CONFOUNDING (SELECTION BIAS)	21	Were the patients in different intervention groups (trials and cohort studies) or were the cases and controls (case-control studies) recruited from the same population?
	22	Were study subjects in different intervention groups (trials and cohort studies) or were the cases and controls (case-control studies) recruited over the same period of time?
	23	Were study subjects randomised to intervention groups?
	24	Was the randomised intervention assignment concealed from both patients and health care staff until recruitment was complete and irrevocable?
	25	Was there adequate adjustment for confounding in the analyses from which the main findings were drawn?
	26	Were losses of patients to follow-up taken into account?
POWER	27	Did the study have sufficient power to detect a clinically important effect where the probability value for a difference being due to chance is less than 5%?

Data Analysis

A descriptive and narrative synthesis approach was used to present the findings from the included studies. This approach was considered appropriate given the heterogeneity of the studies, particularly in the range of study designs and outcome measures used.

Results

Summary of Literature Search

Literature searching took place in several stages following the approach described by Moher, Liberati, Tetzlaff, and Altman (2009) and presented in Figure 1. From the database platforms and other searching, 842 potential studies were identified.

Records from these searches were imported into Endnote X9 and duplicates were removed, leaving 532 studies. Titles and abstracts were reviewed against eligibility criteria resulting in 93 articles remaining for full-text review. These were assessed for eligibility against this review's inclusion criteria. Following this final phase, ten studies were included in the review.

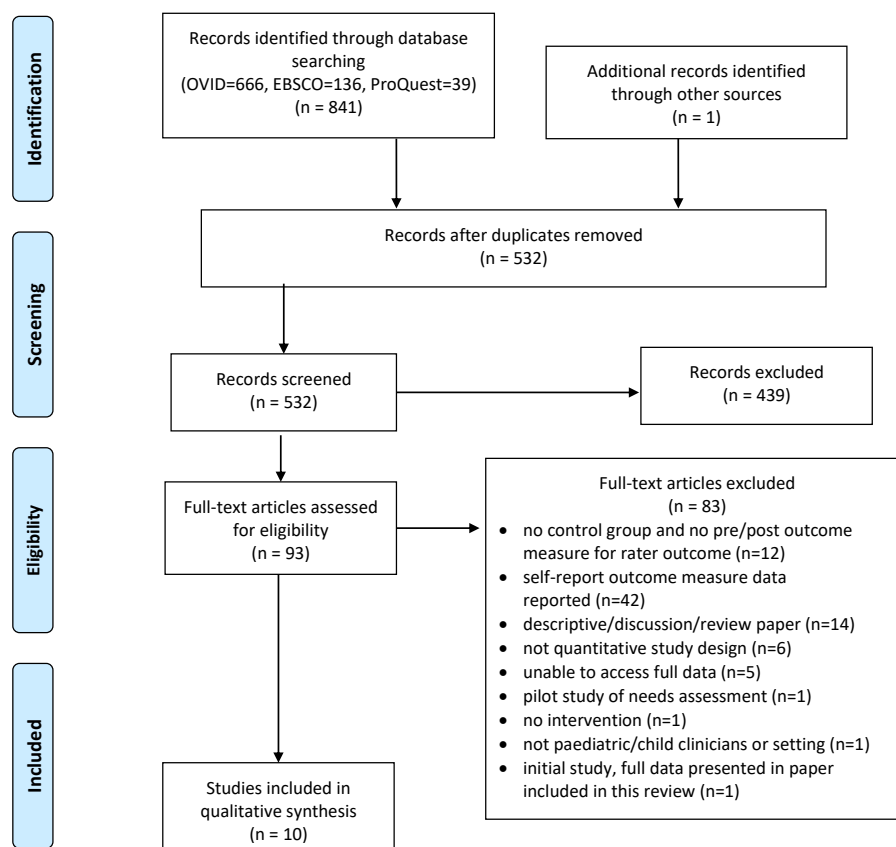


Figure 1 PRISMA flowchart showing process involved in selecting studies, based on Moher et al. (2009)

Overview of included studies

A summary of study characteristics is provided in Table 2. The ten studies included were published in peer-reviewed journals publishing medical research. The large majority of the studies took place in the United States (n=8) (Attar, Hernandez, Mullan, Tang, & Haftel, 2010; Calhoun et al., 2017; Cannone, Atlas, Fornari, Barilla-LaBarca, & Hoffman, 2019; Greenberg, Ochsenschlager, O'Donnell, Mastruserio, & Cohen, 1999; Nellis, Howell, Ching, & Bylund, 2017; Reed et al., 2015; Vaidya, Greenberg, Patel, Strauss, & Pollack, 1999; Yuan, Scott, Van Horn, Oke, & Okada, 2019), one took place in Puerto Rico (Silva, 2008), and another in Canada (Tobler, Grant, & Marczynski, 2014). For ease of reference in the following section, studies will henceforth be referred to by their “study number” allocated in this review (see Table 2 for details).

All studies employed quantitative designs: four were controlled before-after [1,2,5,7], and the remaining six were pre/post designs [3,4,6,8,9,10]. In most studies, participant groups were solely comprised of practitioners working in paediatric or child services (n=7) [1,4,5,7,8,9,10], whilst others included practitioners from a mix of specialty areas (n=3) [2,3,6]. The total sample size was 376. Service settings included in these studies were: paediatric and children's hospital, paediatric emergency care, oncology (including paediatric oncology), palliative care, paediatric intensive care units, and tertiary care children's hospitals and centres.

Table 2: Summary of study characteristics

No.	Primary author, year of publication	Participants	n	Service Setting	Country	Research Design
1	Attar, 2010	Pediatric residents IG: all residents from one cohort CG: residents not exposed to intervention	IG: 23 CG: 11	Pediatrics	United States	Controlled before-after
2	Calhoun, 2017	Emergency Medical Services (EMS) prehospital providers	148 IG:68 CG:80	Pediatric Emergency Medical Service	United States	Controlled before-after
3	Cannone, 2019	Medical trainees and residents	total = 22 (12 in first year, 10 in second year)	Oncology (including pediatric hematology and oncology) and palliative care	United States	Pre and Post Study
4	Greenberg, 1999	2nd and 3rd year pediatric residents and fellows in emergency medicine	27	Children's hospital emergency department	United States	Pre and Post Study
5	Nellis, 2017	Second year pediatric residents	IG: 12 CG: 19	Pediatric Intensive Care Unit	United States	Controlled before-after
6	Reed, 2015	1st year pediatrics and internal medicine/ pediatrics residents	29 (27 pediatrics, 2 medicine/ pediatrics)	Tertiary care children's hospital	United States	Pre and Post Study
7	Silva, 2008	Pediatric residents	IG: 6 CG: 5	Pediatric Tertiary Care Centre	Puerto Rico	Controlled before-after
8	Tobler, 2014	Pediatric trainees (general and emergency)	33	Children's hospital	Canada	Pre and Post Study
9	Vaidya, 1999	Pediatric intensive care fellows	7	Tertiary pediatric intensive care unit in Children's Hospital	United States	Pre and Post Study
10	Yuan, 2019	Pediatric emergency medicine and family medicine residents	34	Pediatric Emergency Medicine Department	United States	Pre and Post Study
Abbreviations: IG: Intervention group, CG: Control group						

All ten review studies examined the effect of interventions on practitioners' ability to communicate bad news and measures of communication were rated by an observer. Some studies included intervention and control groups as well as pre and post measures (n= 4) [1,2,5,7] and those that did not, at least included pre and post measures of communication (n=6) [3,4,6,8,9,10].

Methodological Quality Assessment

Three reviewers (B.G., S.M., and K.G.) individually rated all of the papers using the adapted Downs and Black Checklist items presented in Table 1. As there were more than two raters, Fleiss' kappa was initially considered as an inter-rater reliability statistical measure. However, one of the assumptions (that raters are non-unique) was not met, and Cohen's kappa was therefore used between pairs of raters. On first rating, there was substantial agreement of 89% between B.G. and S.M. (kappa = 0.80, $p < 0.01$), moderate agreement of 73% between K.G. and S.M. (kappa = 0.47, $p < 0.01$), and substantial agreement of 83% between B.G. and K.G. (kappa = 0.64, $p < 0.01$).

Following discussion, any disagreements were resolved and final consensus ratings were given for each criterion. Table 3 provides a summary of the number of papers rated as "yes" or "no/unable to determine" for each criterion, a full list of ratings for each study is presented in Appendix C.

Table 3: Summary of quality assessment by area and criteria based on Downs and Black's Checklist (1998)

Area		Description	Number of studies	
			Rated "yes"	Rated "no" or "unable to determine"
REPORTING	1	Is the hypothesis/aim/ objective of the study clearly described?	8	2
	2	Are the main outcomes to be measured clearly described in the Introduction or Methods section?	10	0
	3	Are the characteristics of the patients included in the study clearly described ?	4	6
	4	Are the interventions of interest clearly described?	10	0
	5	Are the distributions of principal confounders in each group of subjects to be compared clearly described?	4	6
	6	Are the main findings of the study clearly described?	9	1
	7	Does the study provide estimates of the random variability in the data for the main outcomes?	7	3
	8	Have all important adverse events that may be a consequence of the intervention been reported?	2	8
	9	Have the characteristics of patients lost to follow-up been described?	9	1
	10	Have actual probability values been reported (e.g. 0.035 rather than <0.05) for the main outcomes except where the probability value is less than 0.001?	10	0
EXTERNAL VALIDITY	11	Were the subjects asked to participate in the study representative of the entire population from which they were recruited?	3	7
	12	Were those subjects who were prepared to participate representative of the entire population from which they were recruited?	2	8
	13	Were the staff, places, and facilities where the patients were treated, representative of the treatment the majority of patients receive?	10	0
INTERNAL VALIDITY - BIAS	14	Was an attempt made to blind study subjects to the intervention they have received ?	0	10
	15	Was an attempt made to blind those measuring the main outcomes of the intervention?	6	4
	16	If any of the results of the study were based on "data dredging", was this made clear?	9	1
	17	In trials and cohort studies, do the analyses adjust for different lengths of follow-up of patients, or in case-control studies, is the time period between the intervention and outcome the same for cases and controls ?	8	2
	18	Were the statistical tests used to assess the main outcomes appropriate?	10	0
	19	Was compliance with the intervention/s reliable?	9	1
INTERNAL VALIDITY - CONFOUNDING (SELECTION BIAS)	20	Were the main outcome measures used accurate (valid and reliable)?	9	1
	21	Were the patients in different intervention groups (trials and cohort studies) or were the cases and controls (case-control studies) recruited from the same population?	8	2
	22	Were study subjects in different intervention groups (trials and cohort studies) or were the cases and controls (case-control studies) recruited over the same period of time?	8	2
	23	Were study subjects randomised to intervention groups?	3	7
	24	Was the randomised intervention assignment concealed from both patients and health care staff until recruitment was complete and irrevocable?	0	10
	25	Was there adequate adjustment for confounding in the analyses from which the main findings were drawn?	3	7
	26	Were losses of patients to follow-up taken into account?	9	1

Area		Description	Number of studies					
			Size of smallest intervention group					
			<n1	n1-n2	n3-n4	n5-n6	n7-n8	n8+
POWER	27	Did the study have sufficient power to detect a clinically important effect where the probability value for a difference being due to chance is less than 5%?	0	0	0	1	1	8

In the area of reporting, all studies clearly described outcome measures, interventions and reported probability values. Most studies clearly described their aims/hypothesis/objectives (n=8) and their main findings (n=9). The majority provided estimates of random variability (n=7) and described patients lost to follow-up (n=9). A minority of studies described participant characteristics (n=4), and the distributions of principal confounders within groups of participants (n=4). Very few reported all important adverse events that may be a consequence of the intervention (n=2). In terms of external validity, all studies used representative staff, places or facilities whilst only a small number provided sufficient evidence to establish whether subjects and participants were representative of their entire population (n=3) and whether this was the case for those who ended up participating (n=2).

In terms of internal validity relating to bias, all studies were judged to use appropriate statistical tests to assess outcomes. Most appeared to make any “data dredging” clear if this had occurred (n=9) and used the same time period between intervention and outcomes of all participant groups (n=8). The majority used a well described measure (n=9) and suggested compliance with the intervention was reliable (n=9). None of the studies reported blinding subjects to the intervention and just over half attempted to blind those measuring the main outcomes (n=6). Within the area of confounding within internal validity, most studies accounted for losses of participants (n=9) and recruited intervention and controls from the same population (n=8) over the same period of time (n=8). Few studies randomised participants to intervention groups (n=3) or demonstrated adequate adjustment for confounding in analyses (n=3) and no studies could be rated as concealing randomised interventions from both participants and raters. Finally, most studies received the highest power rating

in the checklist (n=8), suggesting they were sufficiently powered to detect a clinically important effect where the probability value for a difference being due to chance is less than 5%.

Overall scores were calculated for each study along with a total percentage using scores from the areas of internal validity, external validity, and power. Percentage scores were used to determine whether each study was categorised as “low”, “moderate” or “high” quality. A summary of the final ratings and quality classifications are presented in Table 4.

Table 4: Summary of rating scores and quality assessment classification by study

Primary author, year of publication	Study no.	Total score (out of 21)	Total Percentage (%)	Descriptive Quality Classification
Attar, 2010	1	14	66.67	Moderate
Cannone, 2019	3	12	57.14	Moderate
Silva, 2018	7	11	52.38	Moderate
Tobler, 2014	8	12	57.14	Moderate
Calhoun, 2017	2	17	80.95	High
Greenberg, 1999	4	15	71.43	High
Nellis, 2014	5	15	71.43	High
Reed, 2017	6	15	71.43	High
Vaidya, 1999	9	17	80.95	High
Yuan, 2019	10	16	76.19	High

No studies were assessed to be within the “low” quality range. Four were rated as being of “moderate” quality [1,3,7,8] and six were assessed as “high” quality [2,4,5,6,9,10]. It is also worth noting that one study, categorised as moderate quality,

had an overall quality score which fell on the border of moderate and high [1].

Overall, these results suggest that the studies included in this review are of good methodological quality, which adds weight to their findings. Examining the interventions and outcome measures used in these studies can therefore provide valid insights into which tools can contribute to improving the communication of bad news in paediatrics settings.

Interventions

For a summary of intervention descriptions, please see Table 5. Interventions were typically administered in a period of hours to a day (n=8) [2,4,5,6,7,8,9,10], with two interventions forming part of a longer-term curriculum spanning eight weeks [3] and three years [1]. Six studies reported a theoretical basis underpinning their intervention [1,2,3,6,7,8]. The SPIKES protocol was most commonly identified (n=4) [1,3,7,8] and was used in all four studies rated as moderate quality. It was used in a modified format incorporating a mindfulness aspect, MR. SPIKES, in one instance [3], alongside two other frameworks: Buckmans' model and HOPE model in another [1] and complemented with adult learning theory models in one more [8]. The remaining theoretical bases for interventions described were the GRIEV_ING Death Notification Protocol [6] and themes acquired through grounded theory analysis of a focus group [2] for two studies assessed to be of high quality.

Role plays or simulations requiring the participants' involvement either with peers, faculty, or standardised patients formed part of the intervention in all but one of the studies [2]. However, the intervention in this paper [2] as well as those in two other

studies [7,8] contained a role play or simulation of breaking bad news performed by others for participants to watch. These were presented either “live” or by video. Therefore, overall, some version of role play/simulation was used in all interventions presented. Within role plays, participants either witnessed or were responsible for delivering bad news related to the health or life of a patient. All interventions used paediatric/child scenarios, either entirely or in combination with adult scenarios.

Didactic teaching related to communicating bad news was a component of the interventions used in the majority of the studies (n=7) [1,2,3,6,7,8,10]. Another component which featured across most of the interventions was an element of debriefing, feedback or reflection (n=9) [1,2,3,4,5,7,8,9,10]. This was led by a range of people across interventions including: more senior healthcare professionals, participants and peers, and standardised patients. Other than the components mentioned above (didactic teaching; role plays or simulations; debriefing, feedback, reflection) some interventions (n=5) also provided participants with additional resources such as a course pack [1], online signposting [2], reading materials [7], and presentation of a specific breaking bad news tool [3,8]. All interventions involved face-to-face contact with peers and one revolved around the use of a specifically-developed smartphone app [2].

The input of an “expert” outside of the research team was reported in just over half of the studies (n=6) [1,2,3,4,8,9]. Three studies involved people relevant to the population being studied (i.e. physicians, paediatric faculty members, healthcare providers) in the development of the intervention or outcome measure used through:

a needs assessment [1,4], piloting and critical appraisal [2], and focus group [2]. Two studies involved colleagues from other disciplines (e.g. psychology, social work) to contribute to intervention delivery [3,8]. Parents who had personal experiences of receiving bad news were reported to be involved in four of the studies [1,2,8,9] and their involvement and contributions varied within these. In one study, the event they had lived through was used as the role play scenario on which residents were evaluated and a videotaped interview of their personal experiences was shown to participants [1]. In another study, parents were part of a focus group used to develop a smartphone app aimed at improving communication of bad news in a study with emergency healthcare providers [2]. In the third study, they were part of the evaluation team assessing paediatric trainees [8] and in the fourth, they formed part of the simulated patient group for role plays [9].

Table 5: Summary of intervention descriptions

No.	Primary author, year of publication	Intervention	Theoretical basis for intervention described	Use of 'expert' outside of research team (e.g. in healthcare area or expert by experience such as parent)
1	Attar, 2010	Curriculum delivered over three years. Each year, residents take part in one hour session including didactic session and peer-peer role play. Those unable to physically attend viewed material using web-based format. In second and third year, residents took lead in CBN to families during neonatal intensive care unit rotations and received feedback on this from supervisors.	Three frameworks: 1. Buckman's model for breaking bad news 2. SPIKES protocol for delivering bad news 3. HOPE model to include spirituality aspect	Residents' needs assessment using pediatric faculty and resident input and practical tips from social worker. CBN scenario based on real case with family. Parents of this case provided experience of receiving bad news via videotaped interview watched by residents following the exercise with SP.
2	Calhoun, 2017	Smartphone app including didactic sessions, videos of simulated breaking bad news events by EMS providers followed by scripted debriefing, signposting to online resources. 3 scenarios were used: a sudden unexplained infant death, motor vehicle accident resulting in death and a suicide. Focus placed on scene management and difficult conversations with families. Voice overs provided explanations and debriefing. IG and CG participated in a simulation role play with SPs in which a infant experiences a cardiac arrest. IG debriefed as a team guided by the app, CG debriefed as "usual". Both took part in another simulated role play 1 to 2 hours later.	Developed from focus group with stakeholders (EMS providers and parents) interpreted using Grounded Theory. Themes identified used as basis of app content.	Focus groups involving 98 EMS providers and 3 parents used to develop smartphone app. App previewed and critically appraised at two conferences and piloted by EMS providers.
3	Cannone, 2019	8 week curriculum, delivered in two hour sessions. Each session addressing different areas of communication. Didactic session with whole group followed by role plays in small groups with faculty, followed by 360 feedback. Topics and role plays following an illness-trajectory (longitudinal course of a cancer patient's illness).	Modified SPIKES protocol: MR. SPIKES	Guest lecturers invited to cover particular topics (psychologist, member of clergy, sex therapist).
4	Greenberg, 1999	Participants take part in simulation in which they communicate bad news to a SP (child death). Following initial simulation, SP provide feedback to participants on their informing and counselling skills. 4 to 10 weeks later, participants take part in a further simulation.	NR	One of outcome measures developed from surveying physicians to determine which skills should be assessed.
5	Nellis, 2017	Participants paired up to take part in simulation with two actors playing SP and nurse. Simulation involved unsuccessful resuscitation of infant. One participant communicated neurological implications of arrest with SP and second participant informed SP of child's death. Participants received structured debriefing and feedback from faculty and SP following simulation. (CG: received informational package of resources focused on communicating with children and families at end of life.) All residents take part in one day bereavement retreat seminar approximately 6 weeks post-simulation and take part in simulation requiring them to deliver bad news to SP.	NR	Unclear

No.	Primary author, year of publication	Intervention	Theoretical basis for intervention described	Use of 'expert' outside of research team (e.g. in healthcare area or expert by experience such as parent)
6	Reed, 2015	Participants trained in "The GRIEV_ING Death Notification Protocol", a 2 hour educational programme including didactic components, small group discussion and role plays. Scenarios specific to pediatrics were used in this intervention. For assessments, participants took part in videotaped encounters with a SP.	The GRIEV_ING Death Notification Protocol	Unclear
7	Silva, 2008	Competency-based workshop consisting of four sessions. Sessions included didactic teaching (based on the "Giving Bad News Checklist"), reading articles (e.g. on SPIKES protocol), viewing videotapes of scenarios in which bad news is delivered appropriately and inappropriately, role plays. Residents received feedback on their performance from SP at different points. 3-4 weeks following workshop sessions, all participants take part in OSCE comprised of two scenarios requiring participants to deliver bad news.	SPIKES protocol	Unclear
8	Tobler, 2014	5 hour simulation-based workshop divided in to two parts. The first included didactic teaching, reflection by participants, presentation of SPIKES tool and observing live role plays of bad news being delivered in a "good" and "bad" manner. The second, was simulation role plays in small groups. 3 scenarios were used and each scenario progresses requiring bad news to be shared with SP at two points: near drowning of infant resulting in death, inflicted brain injury in baby with angry grandparents, and traumatic brain injury in context of parental discord. Participants took part in a group debriefing facilitated by staff member with reflective contributions from peers. Participants took part in videotaped OSCE pre and post intervention in which they were required to give a new diagnosis of either Downs syndrome or leukemia.	SPIKES protocol Adult learning theory (matching workshop content with needs expressed by participants)	Parents who had personal experience of receiving bad news were included in the evaluation of participants. Workshop created by physicians and bereavement social workers.
9	Vaidya, 1999	1 day workshop split in to morning and afternoon sessions. Participants received one of two case scenarios in the morning and the other in the afternoon. Scenarios were of an infant with meningitis and a teenager involved in a motor vehicle accident. Participants were required to take part in role plays with SP breaking bad news to parents and received feedback from SP following first role play.	NR	Some SP selected based on personal experience as a parent.
10	Yuan, 2019	Participants randomly assigned to one of two simulation cases of breaking bad news. One in which an infants is found to have leukemia and the other in which a child has acquired a brain injury following a drowning incident. First, participants take part in role plays with SP. Next they are debriefed and received a didactic teaching session lasting 15-20 minutes covering topics related to breaking bad news. Lastly, participants take part in a further simulation role play and debriefing session, one to two weeks later.	NR	Unclear
Abbreviations: IG: Intervention group, CG: Control group, NR: not reported, CBN: Communicating Bad News, SP: standardised patient/parent, OSCE: Objective Structured Clinical Examination, SPIKES: Summary of framework: Setting Up, Assessing Perception, Invitation to deliver news, Knowledge giving, Empathy, Summary., MR SPIKES: Mindfulness + SPIKES, HOPE: framework incorporating hope, spirituality, religion.				

Outcome Measures

All studies rated communication of breaking bad news using an observer. Observers watched live or recorded role plays of participants in scenarios where they were required to communicate bad news to a simulated patient/parent. In three studies this was done using the Objective Structured Clinical Examination (OSCE) method [3,7,8], while the remaining studies described using either role plays (n=4) [1,6,9,10] or simulations (n=3) [2,4,5]. Summaries of outcome measures and findings are presented in Table 6.

To assess communication, seven studies solely used a pre-existing outcome measure, in either an original or adapted format [1,2,3,6,7,8,10]. The remaining studies used a pre-existing outcome alongside a checklist created by the researchers (n=3) [4,5,9]. Three studies used assessment tools linked to the SPIKES protocol [1,3,7,8] and two used the Patient Perception Questionnaire [4,9]. The following tools were each used by one study: the Gap-Kalamazoo Communication Skills Assessment Form (GKCSAF) [2], the Gibb Trust Scale [4], the Empathic Communication Coding System [5], the GRIEV_ING Death Notification Protocol [6] and the Modified Breaking Bad News Assessment Scale (mBAS) [10], all of which were rated as high quality studies.

Findings

Of the ten studies included in this review, nine found significant differences in communication abilities following intervention [1,2,3,4,6,7,8,9,10], of mostly medium to large effect sizes, with participants demonstrating improvements in skills for

communicating bad news. In a study providing three-month follow-up scores, the improvements remained over time and performance continued to be significantly better than at baseline [6].

Three of the four controlled before and after studies reported pre-intervention comparisons between intervention and control groups [1,2,5] and all found no significant differences in communication between the groups at this point. Post-intervention, statistically significant differences in communication were found between intervention and control groups in three studies, with improved communication ratings in the intervention groups [1,2,7] with large effect sizes. Significant improvements, also with large effect sizes, in communication skills were also reported in intervention group participants between pre and post intervention in these cases. No significant differences in communication between intervention and control group were observed post intervention in one study [5].

In those studies that did not use control groups for comparison but used within-subject pre and post measures for participants, statistically significant differences in areas of communication were observed with mostly medium to large effect sizes. For some, global communication improvements were found (n=4) [3,6,8,9]. Others found improvements in sub-categories (n=2): “communication” and “follow up” skills in a content issues checklist [4], two trust areas (distrust/trust and dependent/interdependent) in the Gibb Trust Scale [4], and the “breaking bad news”, “eliciting concerns”, and “providing information” domains in the Modified Breaking Bad News Assessment Scale (mBAS) [10].

Table 6: Summary of study outcome measures and findings

No.	Primary author, year of publication	Observer rated outcome measure	Measurement time points for observer ratings	Analysis and results	Effect size (Cohen's d)	Findings summary
1	Attar, 2010	Checklist of behaviours used during communication exercise with SP. First year scenario requiring residents to gain consent for an interpreter. Third year requiring residents to CBN to parents that their child had developed an acute and fatal condition.	IG beginning of residency in first year and end of third year of residency CG beginning of residency in first year	Analysis of variance Pre-intervention, IG not significantly different to CG (mean 22.8, sd 3.3 vs mean 20, sd 6.2, p ns) Post-intervention, IG significantly different to CG (mean 26.9, sd 2.1 vs mean 20, sd 6.2, p<0.01) and significantly different from IG baseline (mean 26.9, sd 2.1 vs mean 22.8, sd 3.3, p<0.01)	CG vs IG post intervention [d 1.49] IG pre vs post intervention [d 1.48]	↑ communication skills*
2	Calhoun, 2017	Gap-Kalamazoo Communication Skills Assessment Form (GKCSAF) completed by raters (staff and SP). Checklist containing 9 domains of communication and requiring rater to identify 3 top areas of communication and 3 main areas needing improvement.	During initial simulation and during second simulation (1-2 hours later).	Mann Whitney U Pre intervention, IG not significantly different to CG (mean 2.9, sd 0.58 vs mean 3.0, sd 0.46, p 0.3331) Post intervention, IG significantly different to CG (mean 4.0, sd 0.49 vs mean 3.1, sd 0.50, p<0.001, effect size r 0.69) IG pre intervention significantly different to IG post intervention (mean mean 2.9, sd 0.58 vs mean 4.0, sd 0.49, p<0.001, effect size r 0.69)	CG vs IG post intervention [d 1.82] IG pre vs post intervention [d 2.05]	↑ communication skills using app*
3	Cannone, 2019	1. Videotaped OSCE in which participants were required to break bad news to a SP about a cancer diagnosis. Performance evaluated by faculty using a scoring instrument based on major headings of SPIKES protocol. 2. Checklist completed by SP following each OSCE.	OSCEs (with faculty and SP scoring) before and after completing curriculum.	Paired t-test Faculty Scoring of OSCE Significant positive change with t -3.69 (p 0.0077) SP Scoring of OSCE 1st year significant difference in scores pre and post intervention (mean 26.7, sd 4.88 vs mean 31.5, sd 5.04, p 0.044) 2nd year no significant difference in scores pre and post intervention (mean 38.6, sd 5.93 vs mean 40.9, sd 5.69, p 0.5)	Faculty scoring pre vs post intervention [d -2.61] [^] SP scoring 1st year pre vs post intervention [d 0.97] SP scoring 2nd year pre vs post intervention, ns	↑ global communication proficiency as rated by faculty * ↑ communication skills as rated by SP for 1st year* ↑ communication skills as rated by SP for 2nd year
4	Greenberg, 1999	SP completed checklist to assess content issues (developed from physician survey), Gibb Trust Scale and Patient Perception Questionnaire to assess interpersonal skills	Following first simulation and following second simulation (between 4 and 10 weeks apart).	Wilcoxon signed rank test and paired Student's t test SP ratings Significant difference in 2/5 categories of content issues checklist pre and post intervention "Communication" (mean 6.84, sd 1.07 vs mean 7.38, sd 0.62, p 0.0192) and "Follow-Up" (mean 1.18, sd 0.96 vs mean 1.89, sd 1.36, p 0.0151), no significant difference on "Content", "Support Systems" and "Intervention" categories. Significant difference in 2/4 categories of Gibb Trust Scale pre and post intervention "Distrust/Trust" (p 0.0113) and "Dependent/Interdependent" (p 0.0411).	"Communication" pre vs post intervention [d 0.62] "Follow Up" pre vs post intervention [d 0.60] "Distrust/Trust" pre vs post intervention [d 1.05] [^] "Dependent/Interdependent" pre vs post intervention [d 0.83] [^]	↑ "communication" and "follow up" content skills as rated by SP* ↑ skills in two trust scale areas (distrust/trust and dependent/interdependent)*
5	Nellis, 2014	1. Checklist of communication skills completed by SP and faculty. 2. Empathic Communication Coding System used to assess participants' empathy skills.	Checklist of communication skills and Empathic Communication Coding System completed by SP and faculty following simulations in bereavement retreat (1-4 months post-intervention).	Mann-Whitney test Communication skills checklist scores No significant difference between IG and CG (median 36.3 vs 39.1, p 0.66). Empathic Communication Coding System scores No significant difference between IG and CG (median 4.0 vs 4.1, p 0.59).	ns	No significant differences between IG and CG

No.	Primary author, year of publication	Observer rated outcome measure	Measurement time points for observer ratings	Analysis and results	Effect size (Cohen's d)	Findings summary
6	Reed, 2015	The GRIEV_ING Death Notification Protocol assessment tool examining ten interpersonal and communication competency areas was modified and three subscales were examined "preparation", "bad news delivery", and "wrap-up".	Videotaped encounters with SP used for assessment at three points 1. Baseline - 4 months in to first year of pediatrics training 2. Post educational intervention - 1-2 weeks after educational programme 3. Follow-up - 3 months following intervention	Repeated measures ANOVA <i>Preparation:</i> Significant difference between baseline and post intervention (mean 38.31, sd 12.45 vs mean 67.62, sd 13.11, p<0.001) Significant difference between baseline and follow up (mean 38.31, sd 12.45 vs mean 69.73, sd 12.82, p<0.001) No significant difference between post intervention and follow up (mean 67.62, sd 13.11 vs mean 69.73, sd 12.82, p 0.50) <i>Bad news delivery</i> Significant difference between baseline and post intervention (mean 52.44, sd 12.67 vs mean 73.99, sd 7.93, p<0.001) Significant difference between baseline and follow up (mean 52.44, sd 12.67 vs mean 77.73, sd 7.97, p<0.001) Significant difference between post intervention and follow up (mean 73.99, sd 7.93 vs mean 77.73, sd 7.97, p 0.035) <i>Wrap-Up:</i> Significant difference between baseline and post intervention (mean 18.53, sd 10.09 vs mean 43.97, sd 18.26, p<0.001) Significant difference between baseline and follow up (mean 18.53, sd 10.09 vs mean 42.89, sd 16.93, p<0.001) No significant difference between post intervention and follow up (mean 43.97, sd 18.26 vs mean 42.89, sd 16.93, p 0.76)	Preparation Baseline vs post intervention [d 2.29] Bad news delivery Baseline vs post intervention [d 2.04] Wrap-Up Baseline vs post intervention [d 1.72]	↑ communication performance across three communication subscales* Improvements sustained over time (at 3 month follow up)*
7	Silva, 2008	Giving Bad News Checklist from SPIKES protocol, completed by SP	Both IG and CG were assessed on OSCE performance which took place 3 to 4 weeks following IG completing all 4 workshops sessions.	Mann-Whitney Test Significant difference between intervention and control groups on overall scores across two scenarios (mean 93% vs mean 73%, p 0.001)	IG vs CG [d 2.90]^A	Better skills in giving bad news in those who took part in workshops*
8	Tobler, 2014	Rater evaluation of participant performance using 17 item communication skills checklist	OSCE pre-workshop and OSCE 3 to 6 months post-workshop	Dependent t test Rater evaluation using communication skills checklist Statistical difference between total score pre and post intervention (mean 25.30, sd 5.33 vs mean 27.82, sd 8.33, p 0.004)	Pre vs post intervention [d 0.36]	↑ is communication skills as rated by professionals and parents*
9	Vaidya, 1999	26 item checklist grouped in to 5 categories: communication skills, content issues, support systems, interventions, and the Patient Perception Questionnaire. Checklist completed by rater blinded to order of sessions.	Following each role play one in morning and one in afternoon of workshop day	Paired t-test <i>Overall checklist performance:</i> Significant difference in combined categories score between first and second role play sessions (mean 143.1, SEM(+/-10.0) vs mean 160.8, SEM(+/-10.7) - mean improvement score 18.1 (+/-5.2) p 0.007)	Pre vs post intervention [d 0.57]	↑ improvement in performance of skills for breaking bad news*
10	Yuan, 2019	Modified Breaking Bad News Assessment Scale (mBAS) 22 item checklist containing 5 domains (setting the scene, breaking bad news, eliciting concerns, providing information, general considerations)	Pre teaching intervention (in first role play) and one to two weeks post intervention (during second role play)	Paired t-tests <i>Setting the scene</i> No significant difference between pre and post scores (mean 1.93 vs mean 1.90, mean difference 0.03, sd(+/- 0.65), p .0810) <i>Breaking bad news</i> Significant difference between pre and post scores (mean 2.26 vs mean 1.88, mean difference 0.38, sd (+/-0.81), p 0.013) <i>Eliciting Concerns</i> Significant difference between pre and post scores (mean 2.56 vs mean 2.22, mean difference 0.33, sd (+/-0.60), p 0.004) <i>Providing information</i> Significant difference between pre and post scores (mean 1.98 vs mean 1.68, mean difference 0.30, sd (+/-0.61), p 0.009) <i>General Considerations</i> No significant difference between pre and post scores (mean 1.95 vs mean 1.73, mean difference 0.22, sd (+/-0.63), p 0.054)	Setting the scene ns Breaking bad news Pre vs post intervention [d 0.47] Eliciting Concerns Pre vs post intervention [d 0.55] Providing information Pre vs post intervention [d 0.49] General Considerations ns	↑ communication skills as rated by SP ↑ in 3/5 communication domains (breaking bad news, eliciting concerns and providing information)* ↑ breaking bad news domain*
Abbreviations IG Intervention group, CG Control group, sd standard deviation, * statistically significant, ns not statistically significant, NR not reported, ^A effect size calculated used cited online calculator CBN Communicating Bad News, SP standardised patient/parent, OSCE Objective Structured Clinical Examination, SPIKES Summary of framework Setting Up, Assessing Perception, Invitation to deliver news, Knowledge giving, Empathy, Summary.						

Discussion

This systematic review examined ten studies investigating interventions aimed at improving the communication of bad news in paediatric settings. All studies were assessed to be of either moderate (40%) or high quality (60%) with one study, classified as moderate, being right on the cusp between moderate and high quality. The overall good quality of these studies strengthens the validity of conclusions which can be drawn to assist in answering the review question. Nine of the ten studies reviewed reported significant improvements in communication of bad news following interventions. This suggests that the approaches used are effective in increasing the communication skills assessed in practitioners. The study which did not find significant differences in communication pre and post intervention was assessed as high quality and did not report a theoretical basis for the intervention. It is not, however, possible to determine whether this can account for the findings, as three other interventions from studies also rated as high quality and that also did not report the theoretical basis of their interventions did observe significant improvements in communication skills. Two thirds of the successful interventions that were found to significantly improve communication reported a theoretical basis for their intervention. Given previous findings that interventions following the SPIKES framework resulted in greater improvements in bad news delivery (Johnson & Panagioti, 2018), there is merit in considering the underlying principles upon which interventions are based.

Studies which reported significant improvements in communication shared many features and these were present in studies which received both moderate and high

quality ratings. Role plays or simulations were used in all interventions and the majority also included didactic teaching components. Elements of debriefing, feedback or reflection were common in studies and present in all studies assessed to be high quality which also found significant improvements in communication following intervention. The findings from this review suggest that these features form an important part of this type of training and should be considered in future interventions aiming to improve skills for breaking bad news. This echoes previous findings and recommendations for the use of active and practice-orientated strategies (Berkhof et al., 2011).

Some of the studies reviewed collected participant satisfaction ratings for interventions. These ratings suggest that participants were satisfied and that programmes were well received. This, in combination with the lack of confidence in communication skills reported by participants pre-intervention in some of the review studies and elsewhere in the literature (Dosanjh et al., 2001; Monden et al., 2016; Rider et al., 2008), adds to the evidence that there is a need for this type of training, including from practitioners' perspectives. Interventions were relatively short in duration, lasting between several hours to one day or were incorporated in pre-existing curriculums. As health services need to be mindful of resource and financial costs, short, effective interventions that can be easily implemented need to be considered and may be more appealing in terms of wider roll out.

Variability in communication assessment measures was observed across the studies examined in this review, which poses some difficulty in assessing the effectiveness

of particular interventions in improving communication in a specific area (breaking bad news). Some studies appeared to consider this by choosing a measure that was based on a breaking bad news framework, such as SPIKES, others focused on subscales within pre-existing measures that focused on bad news delivery, and some created specific checklists to try to capture particular skills. Studies rated as higher quality which found significant improvements, tended to use modified measures or measures that had been specifically created for these studies. This may have enabled researchers to focus on particular forms of communication they were hoping to target. There is recognition that validated tools to objectively appraise breaking bad news are limited (e.g. Tobler et al., 2014; Yuan et al., 2019), especially measures looking specifically at this form of communication within paediatric settings. The development and validation of such a tool is an important consideration for future research. This would increase clarity in measuring breaking bad news skills and assessing the effectiveness of interventions, while also facilitating comparisons across studies. Findings from studies using such a tool could be used in meta-analyses, further contributing to this area and strengthening the evidence base for using particular interventions to improve communication of bad news in paediatric settings.

The role of 'experts by experience' parents was highlighted in several of the studies reviewed. Work involving parents in this area enables their experiences to be heard and allows their input to be incorporated in to the development and delivery of interventions. Parents could also play an important role in the creation of appropriate assessment tools, sharing aspects of communication that were important to them. Acknowledging some of the differences in perspectives and priorities that may exist

between parents and healthcare providers (Mack et al., 2005; Muñoz Sastre et al., 2014), working with parents at the level of service evaluation, development, and research will enable more collaboration across multiple levels of healthcare and should continue to be considered in the future.

Of the studies reviewed, one of the higher quality studies included post intervention follow-up measures and found improvements in communication were sustained three months later. There is a lack of data for longer-term follow-ups of communication of bad news skills in paediatrics, which future research could address. This would allow insight into whether skills are retained over greater periods of time. Future work may also look at methods that promote skill retention and training fidelity, such as opportunities for clinicians to engage in additional coaching or supervision sessions. This may mitigate one of the barriers to breaking bad news identified by residents of not feeling emotionally supported (Dosanjh et al., 2001) and possibly reduce stress and burnout whilst increasing skills and confidence.

In paediatrics settings in particular, it is noteworthy that interventions in the studies reviewed here focused on delivering bad news to parents. This review sought to include interventions for communicating bad news to anyone in the child's network but no studies assessed communicating directly with a child. The skill of breaking such news to children and young people, taking into account their understanding and developmental stage, was therefore not addressed. Seeing as these individuals form a core component of paediatric settings, skills in communicating with them are essential. Future studies may need to think carefully about how interventions aimed

at improving these specific skills of communicating bad news to children and young people could be assessed.

The aim of this review was to consider which interventions improve professionals' communication of bad news in paediatric settings. From the studies reviewed here, particular features appeared to contribute to successful interventions. These include the use of: role plays or simulations, didactic teaching, and engagement in forms of feedback or reflection. Interventions may also benefit from input from experts by experience (such as parents) and their assessment may be further strengthened with the development and use of a standardised measure. Future studies could address gaps in certain quality criteria areas that were either not carried out or not reported in the studies reviewed here. Methodological quality of future studies could be improved by ensuring participants are blind to the intervention they are receiving and by using randomised control measured for both participants and raters.

Strengths and limitations

Acknowledging some of the limitations with self-report measures, this review focused on observer ratings of communication skills. It recognised that the designs of research in this area may not take the form of those typically examined in systematic reviews (such as randomised controlled trials) and attempted to capture research from other design methods (such as within-subject pre/post). Whilst this may be a strength in terms of reviewing a broader range of studies, it is important to recognise that these designs may be less robust. During the literature search phase, some possibly relevant research was identified, however it was not possible to access full

data. These pieces of research could be valuable additions and, had they been included, may have influenced the findings presented in the current review. Future reviews could benefit from including these, if access was possible in the future.

Conclusions

This systematic review examined ten quantitative studies assessing interventions to improve the communication of bad news in paediatric settings. Studies were all of moderate or high quality. Significant improvements in communication skills following intervention were found in nine out of the ten studies. Successful interventions shared common features such as role plays or simulations, didactic teaching and elements of debriefing, feedback and reflection. Two-thirds provided a theoretical basis for their intervention. These findings suggest that interventions containing these features can support healthcare professionals in improving their ability to deliver bad news to parents. Future research should consider longer-term follow-ups; the development and implementation of a validated tool to assess breaking bad news skills in paediatrics; the inclusion of parents in service development and research; and interventions to support communicating bad news to children and young people.

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Author information:

Bláthnaid Greene is a trainee clinical psychologist working in the National Health Service specialising in working in the areas of child and adolescent mental health and paediatric psychology.

Mark Hoelterhoff is a lecturer in Clinical Psychology at the University of Edinburgh and a counselling psychologist promoting developing psychological capital in individuals and communities.

Shona Murphy is a clinical psychologist working in paediatric psychology in the National Health Service and is head of the Paediatric Psychology programme with NHS Education for Scotland.

Kara Gibson is a clinical psychologist working in a child and adolescent mental health service in the National Health Service.

References

- Attar, M. A., Hernandez, M. E., Mullan, P. B., Tang, T. S., & Haftel, H. (2010). Pediatric residents' competency in communicating bad news and eliciting spiritual needs: Development and evaluation of a skills-based curriculum. *Journal of Neonatal-Perinatal Medicine*, 3(3), 177-185. doi:10.3233/NPM-2010-0113
- Baile, W. F., Buckman, R., Lenzi, R., Glober, G., Beale, E. A., & Kudelka, A. P. (2000). SPIKES—a six-step protocol for delivering bad news: application to the patient with cancer. *The oncologist*, 5(4), 302-311. doi:10.1634/theoncologist.5-4-302
- Berkhof, M., van Rijssen, H. J., Schellart, A. J., Anema, J. R., & van der Beek, A. J. (2011). Effective training strategies for teaching communication skills to physicians: an overview of systematic reviews. *Patient education and counseling*, 84(2), 152-162. doi:https://doi.org/10.1016/j.pec.2010.06.010
- Brown, R., Dunn, S., Byrnes, K., Morris, R., Heinrich, P., & Shaw, J. (2009). Doctors' stress responses and poor communication performance in simulated bad-news consultations. *Academic Medicine*, 84(11), 1595-1602. doi:10.1097/ACM.0b013e3181baf537
- Buckman, R. (1984). Breaking bad news: why is it still so difficult? *British medical journal (Clinical research ed.)*, 288(6430), 1597. doi:10.1136/bmj.288.6430.1597
- Calhoun, A. W., Sutton, E. R., Barbee, A. P., McClure, B., Bohnert, C., Forest, R., . . . Fallat, M. E. (2017). Compassionate Options for Pediatric EMS (COPE): addressing communication skills. *Prehospital Emergency Care*, 21(3), 334-343. doi:https://doi.org/10.1080/10903127.2016.1263370
- Cannone, D., Atlas, M., Fornari, A., Barilla-LaBarca, M.-L., & Hoffman, M. (2019). Delivering Challenging News: An Illness-Trajectory Communication Curriculum for Multispecialty Oncology Residents and Fellows. *MedEdPORTAL: the journal of teaching and learning resources*, 15. doi:10.15766/mep_2374-8265.10819
- Choudhary, A., & Gupta, V. (2015). Teaching communications skills to medical students: Introducing the fine art of medical practice. *International Journal of*

Applied and Basic Medical Research, 5(Suppl 1), S41. doi:10.4103/2229-516X.162273

- Davies, B., & Connaughty, S. (2002). Pediatric end-of-life care: Lessons learned from parents. *JONA: The Journal of Nursing Administration*, 32(1), 5-6.
- Deeks, J. J., Dinnes, J., D'amico, R., Sowden, A. J., Sakarovitch, C., Song, F., . . . Altman, D. (2003). Evaluating non-randomised intervention studies. *Health technology assessment*, 7(27), 1-179. doi:<https://doi.org/10.3310/hta7270>
- Dosanjh, S., Barnes, J., & Bhandari, M. (2001). Barriers to breaking bad news among medical and surgical residents. *Medical education*, 35(3), 197-205. doi:<https://doi.org/10.1111/j.1365-2923.2001.00766.x>
- Downs, S. H., & Black, N. (1998). The feasibility of creating a checklist for the assessment of the methodological quality both of randomised and non-randomised studies of health care interventions. *Journal of Epidemiology & Community Health*, 52(6), 377-384. doi:<http://dx.doi.org/10.1136/jech.52.6.377>
- Dubé, C. E., LaMonica, A., Boyle, W., Fuller, B., & Burkholder, G. J. (2003). Self-assessment of communication skills preparedness: adult versus pediatric skills. *Ambulatory pediatrics*, 3(3), 137-141. doi:[https://doi.org/10.1367/1539-4409\(2003\)003<0137:SOCSPA>2.0.CO;2](https://doi.org/10.1367/1539-4409(2003)003<0137:SOCSPA>2.0.CO;2)
- Fallowfield, L., & Jenkins, V. (2004). Communicating sad, bad, and difficult news in medicine. *The Lancet*, 363(9405), 312-319. doi:[https://doi.org/10.1016/S0140-6736\(03\)15392-5](https://doi.org/10.1016/S0140-6736(03)15392-5)
- General Medical Council. (2013). *Good Medical Practice*. General Medical Council Retrieved from https://www.gmc-uk.org/-/media/documents/good-medical-practice---english-20200128_pdf-51527435.pdf?la=en&hash=DA1263358CCA88F298785FE2BD7610EB4EE9A530
- Greenberg, L. W., Ochsenschlager, D., O'Donnell, R., Mastruserio, J., & Cohen, G. J. (1999). Communicating bad news: a pediatric department's evaluation of a simulated intervention. *Pediatrics*, 103(6), 1210-1217. doi:<https://doi.org/10.1542/peds.103.6.1210>
- Higgins, J. P., & Green, S. (2011). *Cochrane handbook for systematic reviews of interventions* (Vol. 4): John Wiley & Sons.
- Hulsman, R. L., Pranger, S., Koot, S., Fabriek, M., Karemaker, J. M., & Smets, E. M. (2010). How stressful is doctor–patient communication? Physiological and

- psychological stress of medical students in simulated history taking and bad-news consultations. *International Journal of Psychophysiology*, 77(1), 26-34. doi:<https://doi.org/10.1016/j.ijpsycho.2010.04.001>
- Imo, U. O. (2017). Burnout and psychiatric morbidity among doctors in the UK: a systematic literature review of prevalence and associated factors. *BJPsych bulletin*, 41(4), 197-204. doi:<https://doi.org/10.1192/pb.bp.116.054247>
- James, L., & Johnson, B. (1997). The needs of parents of pediatric oncology patients during the palliative care phase. *Journal of Pediatric Oncology Nursing*, 14(2), 83-95. doi:[https://doi.org/10.1016/S1043-4542\(97\)90006-0](https://doi.org/10.1016/S1043-4542(97)90006-0)
- Johnson, J., & Panagioti, M. (2018). Interventions to Improve the Breaking of Bad or Difficult News by Physicians, Medical Students, and Interns/Residents: A Systematic Review and Meta-Analysis. *Academic Medicine*, 93(9), 1400-1412. doi:10.1097/Acm.0000000000002308
- King, A., & Hoppe, R. B. (2013). "Best practice" for patient-centered communication: a narrative review. *Journal of graduate medical education*, 5(3), 385-393. doi:<https://doi.org/10.4300/JGME-D-13-00072.1>
- Lefebvre, C., Glanville, J., Briscoe, S., Littlewood, A., Marshall, C., Metzendorf, M.-I., . . . Wieland, L. (2019). Chapter 4: Searching for and selecting studies. In T. J. Higgins JPT, Chandler J, Cumpston M, Li T, Page MJ, Welch VA (Ed.), *Cochrane Handbook for Systematic Reviews of Interventions version 6.0 (updated July 2019)*. Cochrane, 2019.
- Levetown, M. (2008). Communicating with children and families: from everyday interactions to skill in conveying distressing information. *Pediatrics*, 121(5), e1441-e1460. doi:<https://doi.org/10.1542/peds.2008-0565>
- Mack, J. W., Hilden, J. M., Watterson, J., Moore, C., Turner, B., Grier, H. E., . . . Wolfe, J. (2005). Parent and physician perspectives on quality of care at the end of life in children with cancer. *J Clin Oncol*, 23(36), 9155-9161. doi:10.1200/JCO.2005.04.010
- Meyer, E. C., Ritholz, M. D., Burns, J. P., & Truog, R. D. (2006). Improving the quality of end-of-life care in the pediatric intensive care unit: parents' priorities and recommendations. *Pediatrics*, 117(3), 649-657. doi:<https://doi.org/10.1542/peds.2005-0144>
- Moher, D., Liberati, A., Tetzlaff, J., & Altman, D. G. (2009). Preferred reporting items for systematic reviews and meta-analyses: the PRISMA statement. *Annals of*

- internal medicine*, 151(4), 264-269. doi:10.7326/0003-4819-151-4-200908180-00135
- Monden, K. R., Gentry, L., & Cox, T. R. (2016). Delivering bad news to patients. *Baylor University Medical Center Proceedings*, 29(1), 101-102. doi:<https://doi.org/10.1080/08998280.2016.11929380>
- Muñoz Sastre, M., Sorum, P., & Mullet, E. (2014). Lay people's and health professionals' views about breaking bad news to children. *Child: care, health and development*, 40(1), 106-114. doi:<https://doi.org/10.1111/j.1365-2214.2012.01420.x>
- Narayanan, V., Bista, B., & Koshy, C. (2010). 'BREAKS' protocol for breaking bad news. *Indian journal of palliative care*, 16(2), 61. doi:10.4103/0973-1075.68401
- National Institute for Health and Clinical Excellence. (2012). *Patient Experience in Adult NHS Services: Improving the Experience of Care for People Using Adult NHS Services (CG138)*. London, UK: National Institute for Health and Care Excellence Retrieved from <https://www.ncbi.nlm.nih.gov/books/NBK115230/>
- Nellis, M. E., Howell, J. D., Ching, K., & Bylund, C. (2017). The use of simulation to improve resident communication and personal experience at end-of-life care. *Journal of pediatric intensive care*, 6(02), 091-097. doi:10.1055/s-0036-1584684
- Nunn, K. (2019). Delivering bad news. *Journal of paediatrics and child health*, 55(6), 617-620. doi:<https://doi.org/10.1111/jpc.14443>
- Ptacek, J., & Eberhardt, T. L. (1996). Breaking bad news: a review of the literature. *Jama*, 276(6), 496-502. doi:10.1001/jama.1996.03540060072041
- Ptacek, J., & McIntosh, E. G. (2009). Physician challenges in communicating bad news. *Journal of behavioral medicine*, 32(4), 380-387. doi:<https://doi.org/10.1007/s10865-009-9213-8>
- Rabow, M. W., & McPhee, S. J. (1999). Beyond breaking bad news: how to help patients who suffer. *Western Journal of Medicine*, 171(4), 260-263.
- Ratcliffe, E., Pickering, S., McLean, S., & Lewis, J. (2014). Is there a relationship between subacromial impingement syndrome and scapular orientation? A systematic review. *Br J Sports Med*, 48(16), 1251-1256. doi:10.1136/bjsports-2013-092389corr1

- Reed, S., Kassis, K., Nagel, R., Verbeck, N., Mahan, J. D., & Shell, R. (2015). Breaking bad news is a teachable skill in pediatric residents: A feasibility study of an educational intervention. *Patient education and counseling*, 98(6), 748-752. doi:<https://doi.org/10.1016/j.pec.2015.02.015>
- Rider, E. A., Volkan, K., & Hafler, J. P. (2008). Pediatric residents' perceptions of communication competencies: Implications for teaching. *Medical teacher*, 30(7), e208-e217. doi:<https://doi.org/10.1080/01421590802208842>
- Shaw, J., Brown, R., Heinrich, P., & Dunn, S. (2013). Doctors' experience of stress during simulated bad news consultations. *Patient education and counseling*, 93(2), 203-208. doi:<https://doi.org/10.1016/j.pec.2013.06.009>
- Shaw, W. S., Zaia, A., Pransky, G., Winters, T., & Patterson, W. B. (2005). Perceptions of provider communication and patient satisfaction for treatment of acute low back pain. *Journal of Occupational and Environmental Medicine*, 47(10), 1036-1043. doi:10.1097/01.jom.0000172863.26222.14
- Silva, D. H. (2008). A competency-based communication skills workshop series for pediatric residents. *B LETÍN*, 8.
- Stewart, M., Meredith, L., Brown, J. B., & Galajda, J. (2000). The influence of older patient-physician communication on health and health-related outcomes. *Clinics in geriatric medicine*, 16(1), 25-36, vii-viii. doi:10.1016/s0749-0690(05)70005-7
- Street Jr, R. L., Makoul, G., Arora, N. K., & Epstein, R. M. (2009). How does communication heal? Pathways linking clinician–patient communication to health outcomes. *Patient education and counseling*, 74(3), 295-301. doi:<https://doi.org/10.1016/j.pec.2008.11.015>
- Street, R. L. (1991). Physicians' communication and parents' evaluations of pediatric consultations. *Medical care*, 29(11), 1146-1152.
- Tobler, K., Grant, E., & Marczinski, C. (2014). Evaluation of the impact of a simulation-enhanced breaking bad news workshop in pediatrics. *Simulation in Healthcare*, 9(4), 213-219. doi:10.1097/SIH.0000000000000031
- Tools, N. C. C. f. M. a. (2008). Quality checklist for health care intervention studies.
- Vaidya, V. U., Greenberg, L. W., Patel, K. M., Strauss, L. H., & Pollack, M. M. (1999). Teaching physicians how to break bad news: a 1-day workshop using standardized parents. *Archives of pediatrics & adolescent medicine*, 153(4), 419-422. doi:10.1001/archpedi.153.4.419

- Wanzer, M. B., Booth-Butterfield, M., & Gruber, K. (2004). Perceptions of health care providers' communication: relationships between patient-centered communication and satisfaction. *Health communication, 16*(3), 363-384.
doi:https://doi.org/10.1207/S15327027HC1603_6
- Wilson, B. (2020, December 1). Practical Meta-Analysis Effect Size Calculator.
<https://campbellcollaboration.org/escalc/html/EffectSizeCalculator-SMD3.php>
- Yuan, Y. Y., Scott, S., Van Horn, N., Oke, O., & Okada, P. (2019). Objective evaluation of a simulation course for residents in the pediatric emergency medicine department: breaking bad news. *Cureus, 11*(1).
doi:10.7759/cureus.3903

Parental experiences of receiving a newborn diagnosis of cystic fibrosis.

Bláthnaid Greene^{1,2,*}, Shona Murphy¹, Mark Hoelterhoff² and Corinne Reid²

¹Psychology Department, NHS Fife, Dunfermline, UK

²School of Health in Social Science, University of Edinburgh, Edinburgh, UK

*Corresponding author: Bláthnaid Greene, contact email: blathnaidgreene@nhs.net

Running head: Parental experiences of newborn diagnosis of CF

^aThis article is written in accordance with author guidelines for Journal of Genetic Counselling (Appendix D). Minor formatting changes were made to provide consistency in this thesis portfolio.

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Abstract

Receiving the diagnosis of cystic fibrosis (CF) is a significant life event for families. With the screening of CF included in newborn screening, families can receive the diagnosis shortly after birth, a time already recognised as bringing significant change and possible challenges. It is important to understand parents' experiences of newborn diagnosis of CF as this can help inform future service delivery. Using Interpretative Phenomenological Analysis, the current study interviewed seven mothers about their experiences of receiving a newborn diagnosis of CF. Interviews were transcribed and analysed with three superordinate themes emerging: Cognitive and Emotional Experiences; Connection; and Knowledge. These results are discussed in relation to existing literature, theories and research. Clinical implications and future research are considered.

Keywords: *cystic fibrosis, newborn screening, diagnosis, parents*

Introduction

Cystic fibrosis (CF) is a life-limiting, genetically inherited condition affecting over 10,500 people in the United Kingdom (Cystic Fibrosis Trust, 2020) and around 900 in Scotland (Scottish Government, 2019). Significant improvements in survival have been observed since the introduction of specialised CF care (Smyth et al., 2014). This includes the implementation of newborn heel prick blood spot screening which covers screening for CF as well as other hereditary conditions (National Health Service, 2018). Newborn screening (NBS) for CF began in Scotland in 2003 (Cystic Fibrosis Trust, 2020).

Chronic health conditions can increase the risk of emotional distress throughout the family (Holmes & Deb, 2003) and the impact on caregiver wellbeing has been highlighted (e.g. Chow, Morrow, Robbins, & Leask, 2013; Cousino & Hazen, 2013). Systematically reviewing literature from the preceding two decades, Berge and Patterson (2004) report that as well as impacts on the individual with CF, psychosocial impacts have been found in siblings and parent caregivers. In a multi-country epidemiological study, parents of children with CF were found to have anxiety and depression rates that were two to three times more elevated than community samples (Quittner et al., 2014). Additionally, adolescents were found to be significantly more likely to be above clinical cut-offs for anxiety and depression if their parents' responses were elevated. Parents of children with CF are required to take on additional caregiving roles such as treatment administration and management which can come with a range of financial, practical and emotional challenges (Fitzgerald, George, Somerville, Linnane, & Fitzpatrick, 2018). These new roles may result in a change in

personal identity for parents and an increase in “burden of responsibility” (Hodgkinson & Lester, 2002), which often occur alongside uncertainty and changes in children’s health status (Fitzgerald et al., 2018). It is therefore important to consider the health and needs of parents of children diagnosed with CF.

Benefits linked to neonatal screening for CF include reduction in disease severity, burden of care and costs (Castellani et al., 2009; Southern, Merelle, Dankert-Roelse, & Nagelkerke, 2009), and increases in quality of life and life expectancy (Smyth et al., 2014). Studies with families who have gone through the screening process report increased levels of anxiety and distress, and though this may be transient depending on the outcome of screening, the experience appears to be common regardless of outcome (Hayeems et al., 2016; Parsons & Bradley, 2003; Ulph, Cullinan, Qureshi, & Kai, 2015). Despite this, parents’ favourable attitudes to newborn screening have been taken as an indicator of the acceptability of early diagnosis (Parsons & Bradley, 2003).

Receiving the news that your child has CF is the beginning of a long-term process and appears to be a salient memory for parents (Chudleigh et al., 2016; Havermans, Tack, Vertommen, Proesmans, & de Boeck, 2015). Using a grounded theory approach to explore experiences of parents receiving a positive NBS result for CF or sickle cell disease, Chudleigh et al. (2016) proposed a theoretical framework in which “complex interactions between factors that occur before, during and after diagnosis” (p.1222) influence parental experiences. Parental understanding of NBS and its possible outcomes, method of diagnosis delivery, and the person communicating this were highlighted as important, as was the need to support parents in sharing news with

others. Queries arose regarding whether diagnosis should be by telephone or in person, though there appeared to be consensus that this should be done by a professional knowledgeable in the condition. Diagnosis is recognised as an emotional time for parents with shock and disbelief reported (Chudleigh et al., 2016; Jessup, Douglas, Priddis, Branch-Smith, & Shields, 2016). In both these studies, parents spoke of how they received and sought information. Considerable variability arose in parents' preferences for the amount, timing and staggering of information presented (Jessup et al., 2016). This variance may reflect the complex processes involved in receiving a diagnosis.

Receiving the diagnosis of CF is a significant life event for families and may influence how they adjust. Diagnosis from NBS occurs during the perinatal period, already a time of 'unparalleled change' for families (Howard, Piot, & Stein, 2014). The current study aims to add to the small body of research in this area by exploring experiences of parents who have received a newborn diagnosis in Scotland, and by considering families' contexts prior to diagnosis. The primary question of this study is "What are parents' experiences of receiving a newborn diagnosis of CF?", aiming to capture how parents experienced the process of their child receiving a CF diagnosis following neonatal screening. To allow a detailed exploration of the rich descriptions of experience provided by parents, it will employ an Interpretative Phenomenological Analysis (IPA) approach. As IPA is "concerned with the detailed examination of personal lived experience, the meaning of experience to participants and how participants make sense of that experience" (Smith, 2011, p. 9) it is viewed as suitable for this study. IPA focuses on describing, understanding and interpreting participants' experiences (Tuohy, Cooney, Dowling, Murphy, & Sixsmith, 2013), not seeking to

provide one 'truth' but to make sense of depictions of experiences. Input and perspectives from service users, including parents, is integral in helping inform health service delivery. IPA is recognised as a suitable approach in healthcare research given its ability to draw links between the meaning participants attribute to events (Smith, Jarman, & Osborn, 1999), their inner cognitive worlds and biopsychological principles used in healthcare (Biggerstaff & Thompson, 2008). As it is interested in providing detailed insight into the personal lived experiences of individuals (Tuffour, 2017) it is believed IPA will support the exploration of parental experiences of receiving a newborn diagnosis of CF.

2. Methods

2.1 Design

As this study aimed to explore the *experience* of parents in relation to a specific event, it employed a qualitative phenomenological research design, an approach recognised for capturing the lived experiences of groups of people (Chenail, 2011; Finlay, 2011). Focusing on experiences of receiving a newborn diagnosis of CF, parents were interviewed using a semi-structured questionnaire. Interviews were recorded, transcribed and analysed following IPA guidelines (Smith, 2011).

2.2 Participants

Seven mothers whose children received a diagnosis of CF from NBS in Scotland participated (both mothers and fathers were invited to participate). This sample size is comparable to IPA studies of parental experiences (Carpenter et al., 2018; Iversen, Esbjørn, Christensen, & Hansen, 2012). Participants met the following criteria: parent of child with CF; adult aged 18 years or older; able to provide

informed consent; diagnosis received from NBS in Scotland; diagnosis received at least six months prior to interview; participant had remained the primary caregiver.

2.3 Recruitment

Participants were recruited through specialist paediatric CF services. CF nurses identified families meeting inclusion criteria and provided those who were interested with participant information sheets (Appendix H). The researcher (B.G.) made contact with those wishing to take part and offered to meet participants in their preferred location: either at home or in clinic. Informed consent (Consent form, Appendix J) was sought after any additional questions were answered and before interviews began. Confidentiality and its limits were made clear and participants were informed that participation was voluntary and they could withdraw at any point.

2.4 Data collection

Participants answered demographic questions and engaged in a 30-60 minute interview. These were guided by a semi-structured questionnaire (Appendix L) which was reviewed by an expert-by-experience parent, CF nurses and a parent advocacy charity. Following IPA approaches (Smith, Flowers, & Larkin, 2009) questions were open-ended and used flexibly to enable parents to discuss issues of importance. Recognising that bridging research and clinical practise is a strength in phenomenological approaches (Finlay, 2011), clinical skills including active listening, reflection, and monitoring participant affect were used. All interviews were recorded on NHS digital recorders. Due to the relatively small parent population, demographic

information is not reported and pseudonyms have been assigned to ensure anonymity.

2.5 Analysis

Given the aim to collect in-depth data regarding the phenomenon of diagnosis, IPA was believed to be a suitable form of analysis to enhance understanding through theme generation. Smith et al.'s (2009) process of analysis was followed. Interviews were transcribed, read and re-read whilst listening to recordings. Initial note-taking was done whilst attempting to "maintain an open mind and note anything of interest within the transcript" (p.83). Following this period of engagement and immersion in the material, emergent themes were identified within interviews. Next, these were grouped to form superordinate themes (Appendix N) and patterns across all interviews were explored. Triangulation methods (Carter, Bryant-Lukosius, DiCenso, Blythe, & Neville, 2014; Patton, 1999) were used to help increase validity and credibility: development of themes was reviewed in supervision, participants provided feedback on data interpretation, and themes were linked to existing theories and findings.

2.6 Reflexivity

Transparency and reflexivity are important in increasing quality in qualitative research (Clancy, 2013). The primary author recognises that certain prejudices as a white, female, middle-class, trainee clinical psychologist, non-parent may affect understanding of results. Though separate from CF teams, the primary author is linked to these healthcare professionals and this may have impacted parents'

account and in turn, the researcher's understanding. Supervision, reflection on the process with colleagues and keeping a reflective journal (Appendix O) were methods used to reduce bias by increasing awareness of attitudes and beliefs held and to promote openness.

2.7 Ethical considerations

This study received ethical approval from the NHS South East Scotland Research Ethics Committee 02 and two health boards (Appendices E, F, G). It was recognised that, given the nature of the topic being explored, interviews may be emotive for participants. The researcher is experienced in working with people who may show signs of distress and was able to monitor appropriately for this. Risk management plans were in place though did not need to be used. A protective factor for participants was existing relationships with CF teams who could provide support if needed. Additional supports were highlighted as part of debriefing (Appendix K).

3. Results

Seven mothers took part in interviews which lasted between 30 and 60 minutes.

From the analysis described above, three superordinate themes, each containing four subordinate themes, emerged. Superordinate themes were labelled: “Cognitive and Emotional Experiences”; “Connection”; and “Knowledge”. Each transcript provided evidence of all three superordinate themes and the large majority provided evidence for each of the subthemes (see Table 1 for a summary of themes by participant). Six of the seven participants provided feedback following their interviews, and all were in agreement that interpretations accurately reflected their experiences.

3.1 Theme 1: Cognitive and Emotional Experiences

Cognitive and emotional experiences regarding the process of diagnosis were described by all mothers. Mothers reported having very specific memories from the time of diagnosis and shared the psychological and emotional impact the diagnosis had on them. They described trying to navigate uncertainty during the diagnosis process and their experiences of making sense of pre-diagnosis difficulties following receiving the news that their child had CF.

3.1.1 Vivid memories

Participants described receiving the diagnosis as a pivotal point of change in their lives, recalling it with clarity. Mothers painted a picture of a life pre-diagnosis that had been ‘typical’ which was then altered when they received the news. Kate pinpointed this shift to the specific moment she received a phone call from professionals:

Kate: "Family life was good, aye life was good... And then the phone rang and that was the changer."

Natalie highlighted her impression of the speed with which the diagnosis changed her experience:

Natalie: "I had to go and change my life all of a sudden from a normal life to a kid that was really needing a lot more help."

As well as clearly recalling the change that came with diagnosis, mothers expressed having very vivid memories from this time. Not only was this evident in the details they provided but many commented explicitly on the quality of their recollections. As Emily shared her recollections, she pointed out being able to remember the exact date. The strong tone with which she communicated this appeared to be a way of emphasising the event's significance, signalling the extent of the impression it left.

Emily: "But then when I got the phone call on the [exact date] – I remember the date – they said "Oh something's come back on his heel prick, someone's gonnae come out to see ya. At 1 o'clock.""

Kate was equally explicit and used a similar tone when sharing the exact time of the diagnosis appointment. Further to this, she used space in the room to physically depict how clearly she could remember where each attendee was sitting.

Kate: "So we went in for the appointment which was half past two. I can still remember that. And [the doctor] was right across from me, his dad was sitting next to me, my mum was next to me [gesturing] and [CF nurse] was in the room."

Participants emphasised the vividness and specificity of their memories from this time, which seems indicative of the significance of their diagnosis experience.

Table 1: Summary of themes by participant

	Kate	Sophie	Natalie	Emily	Sarah	Jane	Grace
Cognitive and Emotional Experiences							
Vivid memories							
Psychological and emotional impact							
Trying to manage uncertainty							
Sense-making with hindsight							
Connection							
To child							
To own social supports							
Through shared experiences							
To CF team							
Knowledge							
Learning development							
Clarity of information							
Seeking hope							
Professionals' attunement							
		= theme identified			= theme not identified in transcript		

3.1.2 Psychological and emotional impact

In recounting their experience of receiving the newborn diagnosis of CF, mothers described the emotional and psychological impact this had on them. Participants' shared feeling in shock when receiving the news and expressed how bad this felt to them. The emotion was palpable during interviews and often resurfaced, as was the case for Emily:

Emily: "Oh horrendous! Absolutely horrendous... yeah the worst day of my life. See even getting upset today just thinking about it. But yeah it was pretty grim like... [starts crying] ...". "It was a very emotional day."

Participants' narratives conveyed the difficulty of receiving the news. The gravity of the situation and the toll this took emotionally and psychologically was expressed by all. Mothers' language reflected this as well as an initial focus on "*all the bad stuff*" (Jane). Natalie likened becoming aware of the diagnosis to experiencing a death:

Natalie: "It was just the whole feeling of it. How do you explain it? It's like somebody had died, it's like somebody has died when you find out. I would have cried for like a week maybe... I'm the type of person that shuts down. I just shut down and stopped talking to everybody that week... It put me in anxiety and like, oh honestly, that week was the horriblest week of my life."".

Experiencing forms of anxiety and lower mood was common amongst participants. Receiving the diagnosis led to certain forms of thinking such as panicking about the future and focusing on negatives and feeling they were in "*a dark place*" (Emily). These forms of thinking impacted on mothers' behaviour and they described experiences of crying or withdrawing in some form.

Kate: "I kind of stopped going out for a while. I mean just even for walks and things. I'd say I probably had a wee bit depression... It was really hard, definitely."

Mothers painted a picture of the experience of diagnosis being a key low point for them. In recounting their levels of distress and emotional reactions participants reflected on this being one of the most challenging points in their journey with CF. Sophie described her mental health being especially affected at this stage and feeling that her family had ended up in unfavourable circumstances:

Sophie: "Like I suffer from anxiety so that was really bad at that point and I was a bit depressed... I did say to her [health visitor] that I was in a real bad, not place, but just like really upset and it wasn't like major depression just sad and crying and just felt like our luck was the worst luck."

3.1.3 Trying to manage uncertainty

Within this difficult process of diagnosis, mothers shared experiencing considerable amounts of uncertainty. They faced into trying to take on board unexpected, and often very unfamiliar, information at a time of shock. At certain points this uncertainty led mothers to be in denial or disbelief:

Sophie: "Emmm it's just not what you want to hear. Just kept on telling myself "She's not got it, she's not got it." Major denial at that point."

Grace recalled grappling with uncertainty regarding her child's health status. She depicted a struggle between disbelief and acceptance:

Grace: "Emmm cos at first I was thinking "nah it's wrong, it's wrong ken its no my bairn". "I did nae want to really, I did nae want to accept it."

With the diagnosis process involving several stages, this uncertainty was often magnified due to families needing to wait between different points of information from professionals or medical tests and procedures. The lack of certainty and clarity appeared to confront participants and perhaps exacerbated their feelings of being out of control. Jane described one example of trying to manage these feelings:

Jane: "... but I had 40 minutes of Google of not having a flipping clue what it could even possibly be so that 40 minutes was actually the worst... Emmm so I think the big things that I look back on that were the worst parts of that day were waiting between the call from the midwife and people arriving because I thought the worst and then probably the delay [in starting medication]."

For participants, it seemed this psychological experience of managing uncertainty formed part of what made receiving the newborn diagnosis so difficult.

3.1.4 Sense-making with hindsight

As well as participants experiencing the diagnosis phase as a period of managing uncertainty it provided an opportunity to make sense of pre-diagnosis symptoms they had been unsure of. With hindsight, mothers were able to better understand previous experiences and difficulties with their babies within the context of CF. This took the form of reviewing the contrast between expectations mothers held about what was 'normal' for babies versus signs their babies were displaying:

Grace: "With [child] it was different to my other kids, because I knew something was wrong... It was dirty bum after dirty bum so it was nae like a normal baby. So I kind of had a feeling something was wrong..."

Hindsight allowed participants to revisit past hypotheses and allowed them to conclude that much of what they had noticed could now be better explained.

Sarah: “And there was wee signs at the start but none of the kids in my family had been breastfed so a lot of things they were saying to me “oh that happens with a breastfed baby”... but at the same time ... there was wee signs.”

Jane: “... in the first few weeks because I had really bad feeding issues like so his weight had dropped. Now everyone attributed that to breastfeeding being a challenge. Probably in hindsight it was because he wasn’t getting the nutrients from the milk, obviously, because of his enzymes.”

Though this, of course, did not make receiving the diagnosis easier, the ability to make sense of past occurrences with hindsight seemed to provide validation for mothers’ earlier feelings that something was ‘wrong’.

Natalie: “... and it was just like when you know something is wrong with the kid. Because I knew something was wrong anyway. The chest wasn’t right and the poos weren’t what you would expect.”

In navigating a different path to the one they had imagined, sense-making with hindsight may have provided reassurance about how well mothers did know their child and their ability to care for them even in the midst of this unexpected diagnosis.

3.2 Theme 2: Connection

Connections to their child, to pre-existing support systems, to new healthcare systems and connections built through shared experiences formed part of participants’ experiences of diagnosis.

3.2.1 To child

Mothers’ narratives demonstrated how they connected to their child with CF. A strong sense of putting their child’s needs first and ensuring they received the care

they needed was echoed in all interviews. Participants' commitment to doing what they felt was best for their child led them to make a range of sacrifices such as ceasing employment, restricting access to previously visited locations due to contamination risks, and continuing to breastfeed despite challenges faced with this.

Kate: "So I guess if he had nae been diagnosed I would have probably just continued to work. But obviously [child] came first so I left..."

Jane: "Because I was like "if this [breastfeeding] helps him, woof! I don't care how sore it is, I will do it"...". "I was focusing on what do I need to do to make him better ... So I was like "OK, well then let me do that [start treatments] now, let me do that as soon as I can because I want him to be as well as possible"."

The connection to their child and drive to care for them appeared to provide participants with additional strength to persevere through difficult circumstances and to act as advocates. Natalie described undergoing a personal transformation:

*Natalie: "Before her I was one of those people, I was like a people pleaser so I never used to want to hurt other people's feelings but since I've had her, I've kind of become, excuse my language, but like a little b***h. So it's just one of those things you have to do and put your foot down whether nobody likes it or not."*

Whilst trying to take on board information in the present during diagnosis, mothers' focus on their child was not only in the current moment but included thinking ahead to their child's future and connecting with what may be in store for their child.

Participants shared their experience of thinking about their child's future health status and the impact CF might have on their life. This led mothers to think about how their child's life might be different to the one they had originally expected for them and losses that may be experienced.

*Sophie: "But in my head, I just kept thinking she's going to be unwell all the time."
"Because you just think "what's my kid's future going to be like?"."*

Mothers' described specific pieces of information standing out and their thoughts seemed to return to these. Particularly salient was the impact CF may have on their child's life expectancy and fertility. On top of the initial diagnosis news, this additional layer of complexity was prominent in mothers' narratives and brought about emotional reactions as they connected with their child's future.

Emily: "So I was like "Oh my God" thinking he wasn't going to be here." "The other thing actually that was soul destroying was when they tell you that they can't have kids, like, themselves."

Jane: "... I genuinely was like "he's gonna die when he's 35, he's not going to be able to have his own family" and genuinely they were the only two things I could think of."

In trying to assimilate unexpected and important information during diagnosis, mothers described a connection to their child across time.

3.2.2 To own social supports

All parents described their experience of social supports during diagnosis. Pre-existing support systems played an important role and mothers explained how they valued this connection with family members in particular. A need to be cared for was expressed and this appeared to be for both emotional and practical support.

Emily: "My mum really, my gran. She's amazing... They were very hands on, they helped loads."

Grace: "If I needed to speak to someone, I'd speak to my sister."

With time to reflect back on this support, mothers shared how crucial they felt it had been to them and commented on how much more difficult they thought things would have been without this connection to their support systems.

Kate: “Don’t know what I would have done without my mum actually. My dad as well... They’re always looking out for us... It’s good to know I’ve got that support there”.

Jane: “I think like I had the immediate support that I needed. I understand that probably a lot of people don’t have that and they did explain to us that there is like a psychology service available but we never actually spoke to anybody because as I said, my husband and I speak when we need to... But also we have amazing family and friends... but I think if you don’t have that support, like if I hadn’t had my husband that day, it would have been horrendous.”

Even where relationships became strained, mothers emphasised the value of receiving support either from that person or another within their trusted group. Kate spoke of this type of connection even within the context of parental separation:

Kate: “We ended up splitting up. And everyone always asks is it because of the pressures. Maybe it is... But I still have a good relationship with him the now. He’s very much supportive... It definitely put a big strain on things.”

3.2.3 Through shared experiences

As well as connecting with specific people, participants described connecting with others through shared experiences. The newborn diagnosis of CF impacted not only parents but those around them as well. Mothers spoke of others sharing in the same emotional reactions.

Natalie: *"I remember one time when I went home everybody started crying."*

Kate: *"Kind of minded it just being a bit like everybody being in shock I think."*

Through these shared experiences, a sense of togetherness emerged as an important part of the diagnosis phase for parents. These seemed to bring people closer together and reinforced mothers' feeling supported by a wider network. Emphasis was placed on the collective experience of receiving the diagnosis and on how others joined parents at this upsetting time.

Emily: *"We were all in bits. My mum, my dad, his dad's parents were there as well. And my sister."*

Connections through shared experiences were also apparent beyond family circles. Others' ability to be empathic, witness, and acknowledge what mothers were going through played a role in promoting connectedness.

Jane [describing CF team]: *"Massively empathetic ... And they did very much listen..." "... she [nurse] obviously wasn't going through it personally per se but, it was like she was like: "I get it, it's awful, it's fine, I'm here" and she gave me a hug again..."*

Sharing experiences joined parents with others. As mothers and others were faced with this difficult news, this connection appeared to provide some comfort.

3.2.4 To CF team

The importance and strength of the relationship between families and the CF team was expressed by all. The diagnostic period is the beginning of a long journey with the team and the start of a process of nurturing a new connection was highlighted in

mothers' narratives. In describing this connection with the CF team, participants expressed how integral the team was in their network, often likening them to family.

Sarah: "It is good because you do form a relationship with them... the stuff you share with them you kinda need to grow a bond."

Emily: "But everything from their part was amazing. You kind of build a friendship with them. They become like your family [laughing]."

Key contributing factors to the success of these connections were the CF team's characteristics and approaches. Their attunement to families and ability to be adaptable and family-centred surfaced across interviews. The team's availability to support families enhanced connections and provided reassurance.

Sarah: "I just have to say the team have been brilliant to me from the start, they really have. Anything at all, I just phone them, there's always somebody there... They've always said: "we're here if you need us, if you need us just phone"."

The flexible support offered by teams, along with a non-judgemental approach, seemed to contribute to parents feeling respected and contained. This enabled families to share honestly and to move at a pace that was better suited to them.

Sophie [about starting treatments]: "But they said: "just one step at a time... just don't rush, when you're ready just start building yourself up... it was good them saying just relax and take it easy."

Grace: "And they were always there, they listened, they would never turn you away if I had a question or was worried about her. The silliest wee things even..."

The value of developing a reciprocal relationship was articulated by mothers and the empathic skills displayed by CF team members contributed to connectedness. This appeared to help empower parents to be active participants in their child's care.

Jane: *"... it was very much like there's a whole team that will help you look after him and even to this day, I feel part of that team... From day one they respond to what we say. Not driven by them or by us, it's a kind of collaboration."*

These characteristics appeared to contribute to the CF teams' ability to provide containment and build trust with families with the term "can't fault them" coming up multiple times. The diagnosis period provided mothers with an opportunity, and a need, to avail of connections already present in their lives and to build new bonds and support systems with their CF teams.

3.3 Theme 3: Knowledge

Across interviews, parents shared the role that knowledge and information played in their experience of diagnosis. Mothers described their experience of knowledge acquisition and development as well as preferences for certain forms of information and knowledge transmission.

3.3.1 Learning development

Common amongst participants was the need to rapidly engage in a new learning process following diagnosis. Six out of seven women had no previous knowledge of CF and many reported being unsure of, or having forgotten, what NBS tested for.

Emily: "I remember him getting the heel prick when he was wee and you don't really even look what it's testing for, you know?" "I had no idea what it [CF] was. I'd never even heard of it before."

Grace: "... but I did nae have a clue cos I'd never heard of any of this."

The one participant who had previous CF knowledge (due to having a relative with CF) acknowledged how she may have felt without this.

Sarah: "[If] I had no experience whatsoever I would have been like "what on Earth?", I would nae even have known what it was if it was not for [relative] to be fair."

The lack of knowledge mothers described having at the time of diagnosis may have added to feelings of distress and shock. During the diagnosis phase, mothers recalled working to establish new routines and learning to administer treatments. Though this was challenging, greater familiarity with these adaptations appeared to increase a sense of control for participants.

Jane: "So yeah, it was new, but within a few days it was just our routine. The hardest thing was probably the night feeds because I was having to ... [gesturing pouring medication, laughing] ... I think it is harder for breastfeeding mums because you don't know how much milk they get."

This development in learning was accompanied by an attitude of needing to "get on with it", often communicated in quite a matter-of-fact tone. Descriptions portrayed this sentiment arising perhaps after a period of assimilating information. This appeared to provide mothers with a way of moving forward for their family.

Grace: "It's the shock more than anything but once you've passed that then you know that the baby needs help ... and you just have to get up and get on with it."

3.3.2 Clarity of information

With receiving and searching for new information forming a critical part of the diagnosis phase, mothers expressed preferences for how this was transmitted to them. All mothers were complimentary of how CF teams shared information with them on the whole. The amount of new information, quantity, and pacing were identified as important factors.

Kate: "I feel like I did get it explained well, they did nae throw everything in one pot at the same time."

Along with getting the right amount of information, a need for the use of clear language was expressed. The emotional responses of participants in combination with trying to take on board unfamiliar information, as described above, may have contributed to mothers seeking language that was easier to comprehend.

Grace: "And they were nae actually telling me what was wrong at first, they were just talking. And big big big words. And I was like looking at my sister like "I dunnae understand what yous are telling me"."

In line with this, participants described appreciating receiving information in an honest and direct form. There seemed to be a recognition that receiving the diagnosis involved hearing difficult information and it was favourable that this was done without trying to minimise aspects.

Emily: "May as well tell everybody how it is. No point in trying to butter it up as such. No, I liked the way, the way they were like "this, this, this and this."."

Wanting clear and reliable information from sources outside of the CF team was also described by participants. Mothers spoke of the need to be cautious when using the internet to search for information based on their experiences and warned of poor quality or inaccurate information.

Sarah: “And the first thing you do is go off and Google it and Google is never going to be a good answer...”

Sophie: “There isn’t really a lot of [good] information. Apart from the CF Trust, Butterfly Trust... You’re best either going on there or asking the CF team themselves...”

Quality and clarity of information were sought by mothers to increase their knowledge and they spoke about the importance of this for families who may receive a diagnosis in the future. This type of information delivery seemed to play an important role in helping families to manage an already challenging time, especially when poor quality information had the potential to add to confusion or distress.

3.3.3 Seeking hope

Alongside a preference for clear information, mothers spoke of including more hopeful or positive facts associated with CF around the time of diagnosis. In emphasising diagnosis as a process, the possibility of pacing or focusing on different parts of the prognosis at certain points in time was raised. Many spoke of starting with issues that needed to be addressed immediately and delaying others aspects.

Jane: “... some of the key bits of information that came out of the first diagnosis that I don’t necessarily think should ... were his life expectancy and the fact that he can’t have his own children...”

It seemed that mothers were trying to manage the weight of the new information that they received during diagnosis and one approach to this was to balance with positive or hopeful information. Participants expressed the benefits of sharing knowledge about research and developments in the field of CF treatments. This was evident when mothers considered future families receiving the diagnosis.

Natalie: "I would say one thing: it's actually not that bad. I know you start thinking "oh it's so bad" but actually it isn't... not to worry so much because there's so much technology out there now..."

Mothers sought hope for their child and the future, some of which they found in acknowledging recent technological developments. It seemed that including information about these developments during diagnosis would be beneficial for families and may provide some comfort during this challenging time.

Jane: "And I think there is so much hope around CF treatment at the minute. And I get it's day one... but none of that was kind of really focused on."

3.3.4 Professionals' attunement

Professionals' ability to be family-centred and adapt to individuals played a key role in mothers' experiences of receiving the diagnosis. On the whole, mothers reported a preference for diagnosis to be delivered by CF professionals, in person (as opposed to by telephone), and within home environments. Mothers' narratives suggest an important factor in successful knowledge transmission was professionals' attunement to families and this seemed to increase mothers feeling contained.

Emily: "So they came to the door and came in. Sat us down and they were amazing. They were really good like. And told us all the stuff and kinda explained in a small way [laughs] what CF is."

Sharing a physical space perhaps facilitated professionals' ability to gauge families' needs and make adaptations accordingly. Mothers valued CF team members' interpersonal skills and appreciated their ability to modify their approach to match individuals.

Jane: *"So I would say the level of information that they give, like in terms of explaining the condition is amazing emm how they adapted to like kind of the different levels of knowledge was really good."*

There was recognition that people may prefer different approaches with the acknowledgment that *"There's no perfect way for a diagnosis..."* (Emily). Instead, what seemed to carry the greatest importance was the need for professionals to pay attention to each family and try to meet their needs as individuals to support them during diagnosis.

Grace: *"But yeah it is, it is trying to figure out what kind of people they are. No like in a bad way just there are different people"*.

This attunement played a key part of participants' experiences of receiving the newborn diagnosis.

Discussion

Using an IPA approach, this study explored the subjective experiences of mothers receiving a newborn diagnosis of cystic fibrosis in Scotland. It is the interviewer's impression that parents were willing and eager to share their experiences. Three superordinate themes emerged from interviews, and all participants who provided

feedback (6/7) reported feeling the interpretations presented here were fair representations of their interview.

Participants in this study all reported cognitive and emotional experiences when sharing their experience of their child receiving a newborn diagnosis of CF. Mothers appeared able to recall this time with ease; and specific details, such as dates, times and sentences used by professionals, were salient in their descriptions. Such recollections may fit with the concept of a particular autobiographical memory known as “flashbulb memories”. These memories are believed to exist for surprising and consequential events (Brown & Kulik, 1977) and have been reported to exist for parents from the time they were first told about their child’s diagnosis of Down’s Syndrome (Ahmed, Ahmed, Jafri, Raashid, & Ahmed, 2015) or of other genetic conditions (Forrest, Curnow, Delatycki, Skene, & Aitken, 2008). In a similar vein, it has been suggested that even one sentence may influence families’ narrative of diagnosis (Tobler, Grant, & Marczinski, 2014). Many parents spoke about the shock experienced in receiving the diagnosis and noted it was a point of significant change from a period of life beforehand that had otherwise been fairly “normal” or “happy”. The importance of this event combined with how unexpected it was could certainly fit the criteria proposed in Brown and Kulik’s flashbulb memory theory.

All mothers described heightened emotions around the diagnosis and shared the impact this had had on their psychological health. The strength of emotions was often visible, with some becoming upset during the interview. These feelings of shock, worry and panic have also been recognised as parental reactions to positive

NBS results for congenital hypothyroidism and CF (Salm, Yetter, & Tluczek, 2012). Experiences of anxiety and low mood fit with the elevated rates of mental health difficulties found in parents of children with CF in general (Besier et al., 2011; Quittner et al., 2014), and may have also been exacerbated at this point as a diagnosis has been identified as a traumatic event (Forrest et al., 2008). Prevalence of post-traumatic stress disorder (PTSD) and post-traumatic stress symptoms have been found to be high in parents of children with CF (Cabizuca et al., 2010). From descriptions provided in interviews, it is possible to imagine how symptoms of post-traumatic stress could exist in this population.

Experiences of connecting with others form a key part of the diagnosis phase for participants. Mothers transmitted a strong sense of holding their child in mind across several domains during these interviews. This moved across time: reflecting back to their child's presentation before diagnosis and, with the benefit of hindsight and increased knowledge, feeling that certain symptoms could now be explained by CF; sharing their child's current health status and how they have managed up until the present day; and thinking ahead to both the nearer future with treatments and developments that may impact them, and times beyond that. It is recognised that parents on the whole think ahead for their children, for example, holding aspirations regarding their future occupations (Irwin & Elley, 2013). In the present study, it seemed that parents' future-thinking included what their child's life may now be like with CF and what challenges they may face. A shortened life expectancy and issues of fertility for those with sons were areas that parents had focused on during diagnosis. In an integrative literature review of 'chronic sorrow' in parents of children with chronic health conditions or disabilities, children's inability or delay in reaching

milestones was found to be associated with parental sorrow (Coughlin & Sethares, 2017). Whilst parents were not discussing developmental milestones that were actively missed in these interviews, it seems that attention was already drawn to what may not be possible for their child in the future. Many spoke of the emotional impact that thinking about this had on them. What surfaced strongly across interviews was parents' drive to prioritise their child, often putting children's needs ahead of their own. The Parent Development Theory posits that parenting is a social and dynamic process, within which parenting is modified over time with the developing child (Mowder, 2005). It proposes that the parenting role comprises six characteristics including general welfare and protection, and responsivity. It may be that for parents in this study, the time of diagnosis led to a modification in their role in response to their child's need for increased care.

The value of empathic connection with others was portrayed through mothers' accounts of receiving support, living shared experiences and building collaborative relationships with their CF team. Parents qualified support from family members as helpful, commenting that they were unsure how they would have managed without this and emphasising the challenges when this support was not available to them. This echoes research which identified social support as a positive coping strategy for parents of a child with CF and found that greater social support seeking was linked to lower emotional impact (Wong & Heriot, 2008). Through describing the reactions and responses of close ones during the diagnosis period, most mothers depicted a shared experience that brought families together. Sharing an emotional experience has been theorised to enhance empathy between the interacting parties, bringing them closer together and as being "instrumental in maintaining, refreshing, and

strengthening important social bonds” (Rimé, 2007, p. 310). The shared emotional experience of diagnosis, often shown by individuals’ reactions to the news, seemed to influence how close or not parents felt towards those individuals.

The concept of strengthening bonds was further emphasised in participants’ portrayals of developing a close relationship with their CF team. Several likened their team to being “your family”; this phenomenon was also described by parents of children with CF living in rural Australia (Jessup et al., 2018). All mothers spoke very positively of their relationships with the teams. Particular characteristics of team members such as being accessible, adaptable, empathic and non-judgemental were emphasised. The importance of practitioner traits in influencing parents’ experience of receiving NBS results has been recognised (Collins et al., 2013) as has the importance of family-centred care (Jessup et al., 2018).

Linked to the idea of parents feeling connected to the teams was professionals’ ability to be attuned to families and providing them with containment. The notion of “containment” (Bion, 1962) is rooted in psychoanalytic approaches and is often described as a process within which one person’s emotional communication can be received by another who is able to ‘hold’ this without becoming overwhelmed, and communicate back to the original person in a way that enables them to feel understood (Douglas, 2007). Much of the way mothers spoke about their interactions with the team resonated with this concept, with one participant connecting the team’s ability to provide her and her husband with containment to their ability to then contain

their own child. Containment has also been recommended as a clinical strategy for working with families with a child with chronic illness (Ødegård, 2005).

The large majority of parents in this study (n=6, 86%) reported having no previous knowledge of CF and many shared an absence of clarity or recollection of what NBS tested for. Lack of knowledge in these areas has been found previously (Chudleigh et al., 2016) with one postal survey study finding 76% of parents of children with CF had not heard of CF before diagnosis (Jedlicka-Köhler, Götz, & Eichler, 1996). As it is acknowledged that NBS takes place during a very busy time for families when they may already be undergoing other procedures and receiving substantial information (Stewart, Hargreaves, & Oliver, 2004), it is perhaps understandable that some information may not be assimilated. The realities of parents grappling with a new phase of life was emphasised in interviews. The transition to parenthood and the learning curve involved in caring for a newborn was described by first time parents as well as those who already had older children. This transition is noted as being an important developmental period for families in general (Deave & Johnson, 2008) which can bring stresses and challenges (Doss, Rhoades, Stanley, & Markman, 2009; Redshaw & Martin, 2014). Trying to manage the diagnosis phase was depicted in interviews as an additional stressor during an already demanding time.

Parents shared how the diagnosis phase brought a period of significant new learning. This learning was more specific to CF than the general newborn learning presented above. Mothers focused on the treatment regimens for their children, particularly administering medication and providing physiotherapy. Adherence to

these is recognised as a complex issue for those with CF and their families (Sawicki, Heller, Demars, & Robinson, 2015) and requires a large amount of input from parents, especially in childhood. The process of learning, including gaining abilities in providing treatments to children, was shared by parents whose child received a diagnosis of Type 1 diabetes (Wennick & Hallström, 2006). Themes from parents interviewed in this Swedish qualitative study suggest that learning occurs at diagnosis and recurs as families face new situations. This is echoed by experiences shared by mothers here who identified specific points of learning (e.g. diagnosis, introduction of new medications) as well as it being an ongoing process. Alongside this new learning was a sentiment of “getting on with it” voiced by many mothers – a sense that parents had no choice but to move ahead. This type of attitude was also observed in a study of caregivers of children with sickle cell disease who described needing to “do what you have to do and move on” despite difficulties (Northington, 2000, p. 149). The authors suggest that this may be a way of having some control whilst having none over the illness itself. It is possible that mothers in the present study may also employ this mentality to exert some control over factors they can actually influence (e.g. tasks of daily life).

The majority of parents received information of the diagnosis face-to-face with the CF team. Those who received it in this manner expressed greater satisfaction with this approach compared to those who received it over the telephone. Overall, parents reported a preference for a diagnosis at home, delivered by CF team members. Parents met with team members, typically a doctor and a nurse, either directly at the diagnosis meeting or face-to-face after the diagnosis when it was given by telephone. Participants recalled the information shared at this encounter

and on the whole reported being pleased with this. Preferences for information to be provided in an upfront and clear manner were voiced and the need for information to be adapted to meet different levels of understanding was acknowledged. As clarity of content message during diagnosis has been linked to parents feeling more positive (e.g. reassured, relieved, calm) (Collins et al., 2013), it could be hypothesised that the pragmatic information transmission sought by these mothers may lead them to feel more reassured as well.

Seeking information from other sources during the diagnosis process seemed to be common amongst mothers. A resource referred to in almost every interview was Google, with mothers warning against googling based on their experiences of finding information that they felt was distressing and sometimes inaccurate or outdated. With increases in the amount of health information online (Ayers & Kronenfeld, 2007) and people's general familiarity with using the Internet, it might be expected that parents would go online for information. Along with paediatricians and physicians, the Internet was found to be one of the most common sources for parents seeking information following a CF diagnosis for their child (Dillard, Shen, Robinson, & Farrell, 2010). Mothers in the present study and elsewhere seemed to use the Internet even when clinicians advised against it (Chudleigh et al., 2016). Possible benefits of Internet-use, such as finding out about advancements and connecting with other parents or individuals with CF, were discussed alongside using websites of charities that had been recommended by CF professionals. The lack of information held by professionals outside of the CF team was discussed by mothers with mostly challenging accounts shared. For many this seemed to add to distressed feelings and a sense of not being supported or contained.

Advancements and developments in the field of CF were discussed by participants. These appeared to provide hope for parents, and some felt they would be important information to share at the time of diagnosis. ‘Vicarious hope’ has been defined as “parent expectations that desirable things will occur in their child’s future” (Wong & Heriot, 2008, p. 345). This has been associated with parental adjustment to CF, being a predictor of child mental health and emotional impact on parents (Wong & Heriot, 2008). This may therefore be an adaptive strategy used by mothers which they felt could benefit others in a similar position. The fact that different people will have different preferences was acknowledged across many interviews with some mothers explicitly stating that there is no perfect way to provide a diagnosis. This concept that there is “no one size fits all” for delivering information is also reflected in the literature (Jessup et al., 2016, p. 240).

Study Strengths and Limitations

A strength of this study is its provision of a detailed analysis of the phenomenon of newborn diagnosis of CF as experienced by mothers. Results presented from the current sample resonate with previous studies. Findings are strengthened by the use of participant feedback which agreed with the themes generated. Whilst IPA enables examination of specific accounts, it focuses on the accounts of the current participants and other narratives may have been missed. As a result, it cannot be assumed that the findings presented here can be generalised to the whole population. The researcher’s subjectivity and active role also needs to be acknowledged, as another researcher may have approached developing the study and conducting interviews differently, leading to different information being gathered.

Areas of focus and interpretations may have been influenced by the experience of the researcher, for example, due to working in the field of clinical psychology, and it is possible that a researcher with a different professional background may have yielded different results.

Recruitment may have been biased by the CF teams who identified participants. This may have led to parents with good pre-existing relationships with CF teams being more likely to be invited to participate. A self-selection bias may also have influenced the findings of this study as specific mothers who wanted or needed to share their story or who may have held particular opinions may have been more likely to take part. It is possible, for example, that the experiences of parents who did not feel able to participate in an interview may have been missed. Knowing that feedback would be provided to CF teams may have also influenced what participants shared. Though attempts were made to recruit from several geographical areas, fewer areas participated in this research than originally anticipated. This means accounts are focused on experiences with a small number CF teams and it cannot be assumed that all findings are generalisable to the whole population. Future larger scale studies, including participants from a greater number of health board areas, may help address these issues and increase the generalisability of findings.

Practice Implications and Research Recommendations

Narratives shared in this study present the psychological and emotional impact of diagnosis on the mothers interviewed. Awareness and acknowledgement of this impact may help professionals better support families and monitor families for

potential distress or mental health difficulties. The value and need for connection with others were apparent in interviews and should continue to be offered, nurtured, and actively monitored for in families. Ensuring families feel adequately connected to networks in their personal lives as well as through healthcare teams may contribute to families feeling contained and better able to cope. Accounts shared in this study suggest a preference for newborn CF diagnosis to be delivered at home, by CF specialists. In line with recognising the great amount of knowledge sharing and acquisition during diagnosis, the value of good communication and interpersonal skills was evident across all accounts. Recognising that no “one size fits all”, the need for professionals to be family-centred and attuned to individuals was highlighted. This requires gauging family contexts and abilities in a short amount of time to then be able to respond appropriately, adapting information and pacing delivery. Skills promoting containment, such as empathy and active listening, will need to be employed to help families feel supported during diagnosis.

The role of online information, and ways to buffer distressing or inaccurate information, will need to be considered by clinicians. Families could be asked explicitly about information they have acquired so any misinformation can be corrected. Teams could provide information digitally if this is the mode increasingly used by families and future research could investigate the effectiveness of different information platforms on increasing accurate parental knowledge. Such developments could benefit from involving parents. Future research could also benefit from looking at fathers’ experiences of diagnosis as this was not captured in the present study. Larger quantitative studies may provide insights into the effect of

certain methods of newborn CF diagnosis delivery on parental satisfaction and wellbeing.

Conclusions

The newborn diagnosis of CF is a period remembered clearly by mothers and is a time accompanied by substantial cognitive and emotional experiences. The value of connection, empathy and the need for support was highlighted. Participants shared the role of connection through pre-existing and new relationships with others as well as through shared experiences at this time. Alongside diagnosis comes substantial new learning and adjustment. Knowledge development forms a key part of this period and information delivery and content needs to be carefully considered by clinicians. Communication and interpersonal skills, such as attunement, play an important role in providing containment to families and impacted the experience of diagnosis as described by the mothers in this study.

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Conflict of Interest

Shona Murphy works as part of one of the CF teams. No conflicts of interests were identified for Bláthnaid Greene, Mark Hoelterhoff or Corinne Reid.

Human Studies and Informed Consent

Ethical approval was granted by the NHS South East Scotland Research Ethics Committee 02, NHS Fife and NHS Lothian (Appendices E, F, G). All participants provided informed consent.

Animal Studies

No non-human animal studies were carried out by the authors for this article.

References

- Ahmed, K. J., Ahmed, M., Jafri, H. S., Raashid, Y., & Ahmed, S. (2015). Pakistani mothers' and fathers' experiences and understandings of the diagnosis of Down syndrome for their child. *Journal of community genetics*, 6(1), 47-53.
doi:<https://doi.org/10.1007/s12687-014-0200-6>
- Ayers, S. L., & Kronenfeld, J. J. (2007). Chronic illness and health-seeking information on the Internet. *Health*, 11(3), 327-347.
doi:<https://doi.org/10.1177/1363459307077547>
- Berge, J. M., & Patterson, J. M. (2004). Cystic fibrosis and the family: A review and critique of the literature. *Families, Systems, & Health*, 22(1), 74.
doi:<https://doi.org/10.1037/1091-7527.22.1.74>
- Besier, T., Born, A., Henrich, G., Hinz, A., Quittner, A. L., Goldbeck, L., & Group, T. S. (2011). Anxiety, depression, and life satisfaction in parents caring for children with cystic fibrosis. *Pediatric pulmonology*, 46(7), 672-682.
doi:<https://doi.org/10.1002/ppul.21423>
- Biggerstaff, D., & Thompson, A. R. (2008). Interpretative phenomenological analysis (IPA): A qualitative methodology of choice in healthcare research. *Qualitative research in psychology*, 5(3), 214-224.
doi:<https://doi.org/10.1080/14780880802314304>
- Bion, W. R. (1962). *Learning from Experience*. . London/New York: Karnac.
- Brown, R., & Kulik, J. (1977). Flashbulb memories. *Cognition*, 5(1), 73-99.
doi:[https://doi.org/10.1016/0010-0277\(77\)90018-X](https://doi.org/10.1016/0010-0277(77)90018-X)
- Cabizuca, M., Mendlowicz, M., Marques-Portella, C., Ragoni, C., Coutinho, E. S. F., de Souza, W., . . . Figueira, I. (2010). The invisible patients: posttraumatic stress disorder in parents of individuals with cystic fibrosis. *Revista de Psiquiatria Clínica*, 37(1), 6-17. Retrieved from
file:///C:/Users/blath/Downloads/17319-Article%20Text-20792-1-10-20120522%20(2).pdf
- Carpenter, K., Wittkowski, A., Hare, D. J., Medford, E., Rust, S., Jones, S. A., & Smith, D. M. (2018). Parenting a child with phenylketonuria (PKU): an interpretative phenomenological analysis (IPA) of the experience of parents. *Journal of genetic counseling*, 27(5), 1074-1086.
doi:<https://doi.org/10.1007/s10897-018-0227-7>

- Carter, N., Bryant-Lukosius, D., DiCenso, A., Blythe, J., & Neville, A. J. (2014). *The use of triangulation in qualitative research*. Paper presented at the Oncology nursing forum.
- Castellani, C., Southern, K. W., Brownlee, K., Roelse, J. D., Duff, A., Farrell, M., . . . Sermet-Gaudelus, I. (2009). European best practice guidelines for cystic fibrosis neonatal screening. *Journal of cystic fibrosis*, 8(3), 153-173. doi:http://doi.org/10.1016/j.jcf.2009.01.004
- Chenail, R. J. (2011). Ten Steps for Conceptualizing and Conducting Qualitative Research Studies in a Pragmatically Curious Manner. *Qualitative Report*, 16(6), 1713-1730. doi:https://files.eric.ed.gov/fulltext/EJ956240.pdf
- Chow, M. Y. K., Morrow, A. M., Robbins, S. C. C., & Leask, J. (2013). Condition-specific quality of life questionnaires for caregivers of children with pediatric conditions: A systematic review. *Quality of Life Research*, 22(8), 2183-2200. doi:https://doi.org/10.1007/s11136-012-0343-z
- Chudleigh, J., Buckingham, S., Dignan, J., O'Driscoll, S., Johnson, K., Rees, D., . . . Metcalfe, A. (2016). Parents' experiences of receiving the initial positive newborn screening (NBS) result for cystic fibrosis and sickle cell disease. *Journal of genetic counseling*, 25(6), 1215-1226. doi:10.1258/jrsm.2008.s18006.
- Clancy, M. (2013). Is reflexivity the key to minimising problems of interpretation in phenomenological research? *Nurse researcher*, 20(6). doi:10.7748/nr2013.07.20.6.12.e1209
- Collins, J. L., La Pean, A., O'Tool, F., Eskra, K. L., Roedl, S. J., Tluczek, A., & Farrell, M. H. (2013). Factors that influence parents' experiences with results disclosure after newborn screening identifies genetic carrier status for cystic fibrosis or sickle cell hemoglobinopathy. *Patient education and counseling*, 90(3), 378-385. doi:10.1016/j.pec.2011.12.007
- Coughlin, M. B., & Sethares, K. A. (2017). Chronic sorrow in parents of children with a chronic illness or disability: An integrative literature review. *Journal of pediatric nursing*, 37, 108-116. doi:https://doi.org/10.1016/j.pedn.2017.06.011
- Cousino, M. K., & Hazen, R. A. (2013). Parenting stress among caregivers of children with chronic illness: a systematic review. *Journal of pediatric psychology*, 38(8), 809-828. doi:http://doi.org/10.1093/jpepsy/jst049

- Cystic Fibrosis Trust. (2020). What is cystic fibrosis? Retrieved from <https://www.cysticfibrosis.org.uk/what-is-cystic-fibrosis>
- Deave, T., & Johnson, D. (2008). The transition to parenthood: what does it mean for fathers? *Journal of advanced nursing*, 63(6), 626-633.
doi:<https://doi.org/10.1111/j.1365-2648.2008.04748.x>
- Dillard, J. P., Shen, L., Robinson, J. D., & Farrell, P. M. (2010). Parental information seeking following a positive newborn screening for cystic fibrosis. *Journal of health communication*, 15(8), 880-894.
doi:<https://doi.org/10.1080/10810730.2010.522226>
- Doss, B. D., Rhoades, G. K., Stanley, S. M., & Markman, H. J. (2009). The effect of the transition to parenthood on relationship quality: an 8-year prospective study. *Journal of personality and social psychology*, 96(3), 601.
doi:<https://doi.org/10.1037/a0013969>
- Douglas, H. (2007). *Containment and reciprocity: Integrating psychoanalytic theory and child development research for work with children*: Routledge.
- Finlay, L. (2011). *Phenomenology for therapists: Researching the lived world*: John Wiley & Sons.
- Fitzgerald, C., George, S., Somerville, R., Linnane, B., & Fitzpatrick, P. (2018). Caregiver burden of parents of young children with cystic fibrosis. *Journal of cystic fibrosis*, 17(1), 125-131. doi:<http://doi.org/10.1016/J.JCF.2017.08.016>
- Forrest, L. E., Curnow, L., Delatycki, M. B., Skene, L., & Aitken, M. (2008). Health first, genetics second: exploring families' experiences of communicating genetic information. *European Journal of Human Genetics*, 16(11), 1329-1335. doi:<https://doi.org/10.1038/ejhg.2008.104>
- Havermans, T., Tack, J., Vertommen, A., Proesmans, M., & de Boeck, K. (2015). Breaking bad news, the diagnosis of cystic fibrosis in childhood. *Journal of cystic fibrosis*, 14(4), 540-546. doi:<http://doi.org/10.1016/j.jcf.2014.12.005>
- Hayeems, R. Z., Miller, F. A., Barg, C. J., Bombard, Y., Kerr, E., Tam, K., . . . Davies, C. (2016). Parent experience with false-positive newborn screening results for cystic fibrosis. *Pediatrics*, 138(3), e20161052. doi:DOI:
<https://doi.org/10.1542/peds.2016-1052>
- Hodgkinson, R., & Lester, H. (2002). Stresses and coping strategies of mothers living with a child with cystic fibrosis: implications for nursing professionals.

- Journal of advanced nursing*, 39(4), 377-383.
doi:<http://doi.org/10.1046/j.1365-2648.2002.02299.x>
- Holmes, A. M., & Deb, P. (2003). The effect of chronic illness on the psychological health of family members. *Journal of Mental Health Policy and Economics*, 6(1), 13-22. Retrieved from <https://pdfs.semanticscholar.org/1896/d37654641eb50ac569786470d20eeaba31b4.pdf>
- Howard, L. M., Piot, P., & Stein, A. (2014). No health without perinatal mental health. *The Lancet*. doi:[http://doi.org/10.1016/S0140-6736\(14\)62040-7](http://doi.org/10.1016/S0140-6736(14)62040-7)
- Irwin, S., & Elley, S. (2013). Parents' hopes and expectations for their children's future occupations. *The Sociological Review*, 61(1), 111-130.
doi:<https://doi.org/10.1111/j.1467-954X.2012.02139.x>
- Iversen, A. E., Esbjørn, B. H., Christensen, E., & Hansen, N. S. (2012). Exploring the impact of involving fathers in the treatment of their children: A study using Interpretative Phenomenological Analysis (IPA). *Qualitative Studies*, 3(1), 22-39. doi:<https://doi.org/10.7146/qs.v3i1.6271>
- Jedlicka-Köhler, I., Götz, M., & Eichler, I. (1996). Parents' recollection of the initial communication of the diagnosis of cystic fibrosis. *Pediatrics*, 97(2), 204-209.
- Jessup, M., Douglas, T., Priddis, L., Branch-Smith, C., & Shields, L. (2016). Parental experience of information and education processes following diagnosis of their infant with cystic fibrosis via newborn screening. *Journal of pediatric nursing*, 31(3), e233-e241. doi:<https://doi.org/10.1016/j.pedn.2015.11.010>
- Jessup, M., Smyth, W., Abernethy, G., Shields, L., Douglas, T., & AREST-CF. (2018). Family-centred care for families living with cystic fibrosis in a rural setting: A qualitative study. *Journal of clinical nursing*, 27(3-4), e590-e599. doi:<https://doi.org/10.1111/jocn.14105>
- Mowder, B. A. (2005). Parent development theory: Understanding parents, parenting perceptions and parenting behaviors. *Journal of Early Childhood and Infant Psychology*, 1, 45-65.
- National Health Service. (2018). Cystic fibrosis. Retrieved from <https://www.nhs.uk/conditions/cystic-fibrosis/>
- Northington, L. (2000). Chronic sorrow in caregivers of school age children with sickle cell disease: A grounded theory approach. *Issues in Comprehensive*

- Pediatric Nursing*, 23(3), 141-154.
doi:<https://doi.org/10.1080/01460860050174693>
- Ødegård, W. (2005). Chronic illness as a challenge to the attachment process. *Clinical child psychology and psychiatry*, 10(1), 13-22.
doi:<https://doi.org/10.1177/1359104505048787>
- Parsons, E. P., & Bradley, D. M. (2003). Psychosocial issues in newborn screening for cystic fibrosis. *Paediatric respiratory reviews*, 4(4), 285-292.
doi:[http://doi.org/10.1016/S1526-0542\(03\)00086-1](http://doi.org/10.1016/S1526-0542(03)00086-1)
- Patton, M. Q. (1999). Enhancing the quality and credibility of qualitative analysis. *Health services research*, 34(5 Pt 2), 1189.
- Quittner, A. L., Goldbeck, L., Abbott, J., Duff, A., Lambrecht, P., Solé, A., . . . Catastini, P. (2014). Prevalence of depression and anxiety in patients with cystic fibrosis and parent caregivers: results of The International Depression Epidemiological Study across nine countries. *Thorax*, 69(12), 1090-1097.
doi:<http://dx.doi.org/10.1136/thoraxjnl-2014-205983>
- Redshaw, M., & Martin, C. (2014). The couple relationship before and during transition to parenthood. doi:<https://doi.org/10.1080/02646838.2014.896146>
- Rimé, B. (2007). The social sharing of emotion as an interface between individual and collective processes in the construction of emotional climates. *Journal of social issues*, 63(2), 307-322. doi:<https://doi.org/10.1111/j.1540-4560.2007.00510.x>
- Salm, N., Yetter, E., & Tluczek, A. (2012). Informing parents about positive newborn screen results: parents' recommendations. *Journal of Child Health Care*, 16(4), 367-381. doi:<https://doi.org/10.1177/1367493512443906>
- Sawicki, G. S., Heller, K. S., Demars, N., & Robinson, W. M. (2015). Motivating adherence among adolescents with cystic fibrosis: youth and parent perspectives. *Pediatric pulmonology*, 50(2), 127-136. doi:
<https://doi.org/10.1002/ppul.23017>
- Scottish Government. (2019). Health Secretary welcomes cystic fibrosis drug pricing agreement. Retrieved from <https://www.gov.scot/news/health-secretary-welcomes-cystic-fibrosis-drug-pricing-agreement/>
- Smith, J., Flowers, P., & Larkin, M. (2009). Planning an IPA research study. *Interpretative phenomenological analysis: theory, method and research*. London: SAGE Publications, 40-55.

- Smith, J. A. (2011). Evaluating the contribution of interpretative phenomenological analysis: A reply to the commentaries and further development of criteria. *Health psychology review*, 5(1), 55-61.
doi:<http://doi.org/10.1080/17437199.2010.510659>
- Smith, J. A., Jarman, M., & Osborn, M. (1999). Doing interpretative phenomenological analysis. *Qualitative health psychology: Theories and methods*, 218-240.
- Smyth, A. R., Bell, S. C., Bojcin, S., Bryon, M., Duff, A., Flume, P., . . . Schwarzenberg, S. J. (2014). European cystic fibrosis society standards of care: best practice guidelines. *Journal of cystic fibrosis*, 13, S23-S42.
doi:<https://doi.org/10.1016/j.jcf.2014.03.010>
- Southern, K. W., Merelle, M. M., Dankert-Roelse, J. E., & Nagelkerke, A. (2009). Newborn screening for cystic fibrosis. *Cochrane Database of Systematic Reviews*(1). doi:<http://doi.org/10.1002/14651858.CD001402.pub2>
- Stewart, R., Hargreaves, K., & Oliver, S. (2004). Telling parents about the heel-prick test: consultation results and resource development. *London: UK Newborn Screening Programme Centre*. Retrieved from https://www.researchgate.net/profile/Ruth_Stewart/publication/42385256_Telling_parents_about_the_heel-prick_test_consultation_results_and_resources_development/links/00b7d513f633ac8f57000000.pdf
- Tobler, K., Grant, E., & Marczyński, C. (2014). Evaluation of the impact of a simulation-enhanced breaking bad news workshop in pediatrics. *Simulation in Healthcare*, 9(4), 213-219. doi:10.1097/SIH.0000000000000031.
- Tuffour, I. (2017). A critical overview of interpretative phenomenological analysis: a contemporary qualitative research approach. *Journal of Healthcare Communications*, 2(4), 52. doi:DOI: 10.4172/2472-1654.100093
- Tuohy, D., Cooney, A., Dowling, M., Murphy, K., & Sixsmith, J. (2013). An overview of interpretive phenomenology as a research methodology. *Nurse researcher*, 20(6). doi:10.7748/nr2013.07.20.6.17.e315
- Ulph, F., Cullinan, T., Qureshi, N., & Kai, J. (2015). Parents' responses to receiving sickle cell or cystic fibrosis carrier results for their child following newborn screening. *European Journal of Human Genetics*, 23(4), 459-465.
doi:<http://doi.org/10.1038/ejhg.2014.126>

Wennick, A., & Hallström, I. (2006). Swedish families' lived experience when a child is first diagnosed as having insulin-dependent diabetes mellitus: an ongoing learning process. *Journal of Family Nursing*, 12(4), 368-389.

doi:<https://doi.org/10.1177/1074840706296724>

Wong, M., & Heriot, S. (2008). Parents of children with cystic fibrosis: how they hope, cope and despair. *Child: care, health and development*, 34(3), 344-354.

doi: <https://doi.org/10.1111/j.1365-2214.2007.00804.x>

Appendix A – Author guidelines for Journal of Communication in Healthcare

Instructions for authors

Thank you for choosing to submit your paper to us. These instructions will ensure we have everything required so your paper can move through peer review, production and publication smoothly. Please take the time to read and follow them as closely as possible, as doing so will ensure your paper matches the journal's requirements.

This title utilises format-free submission. Authors may submit their paper in any scholarly format or layout. References can be in any style or format, so long as a consistent scholarly citation format is applied.

About the Journal

Journal of Communication in Healthcare is an international, peer-reviewed journal publishing high-quality, original research. Please see the journal's Aims & Scope for information about its focus and peer-review policy.

Please note that this journal only publishes manuscripts in English.

Journal of Communication in Healthcare accepts the following types of article: original articles and patient voices.

Peer Review and Ethics

Taylor & Francis is committed to peer-review integrity and upholding the highest standards of review. Once your paper has been assessed for suitability by the editor, it will then be double blind peer reviewed by independent, anonymous expert referees. Find out more about what to expect during peer review and read our guidance on publishing ethics.

Preparing Your Paper

All authors submitting to medicine, biomedicine, health sciences, allied and public health journals should conform to the Uniform Requirements for Manuscripts Submitted to Biomedical Journals, prepared by the International Committee of Medical Journal Editors (ICMJE).

Structure

Your paper should be compiled in the following order: title page; abstract; keywords; main text introduction, materials and methods, results, discussion; acknowledgments; declaration of interest statement; references; appendices (as appropriate); table(s) with caption(s) (on individual pages); figures; figure captions (as a list).

Word Limits

Please include a word count for your paper.

A typical paper for this journal should be no more than 6500 words.

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Authors may submit their paper in any scholarly format or layout. Manuscripts may be supplied as single or multiple files. These can be Word, rich text format (rtf), open document format (odt), or PDF files. Figures and tables can be placed within the text or submitted as separate documents. Figures should be of sufficient resolution to enable refereeing.

- There are no strict formatting requirements, but all manuscripts must contain the essential elements needed to evaluate a manuscript: abstract, author affiliation, figures, tables, funder information, and references. Further details may be requested upon acceptance.
- References can be in any style or format, so long as a consistent scholarly citation format is applied. Author name(s), journal or book title, article or chapter title, year of publication, volume and issue (where appropriate) and page numbers are essential. All bibliographic entries must contain a corresponding in-text citation. The addition of DOI (Digital Object Identifier) numbers is recommended but not essential.
- The journal reference style will be applied to the paper post-acceptance by Taylor & Francis.
- Spelling can be US or UK English so long as usage is consistent.

Note that, regardless of the file format of the original submission, an editable version of the article must be supplied at the revision stage.

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Checklist: What to Include

1. **Author details.** Please ensure everyone meeting the International Committee of Medical Journal Editors (ICMJE) requirements for authorship is included as an author of your paper. All authors of a manuscript should include their full name and affiliation on the cover page of the manuscript. Where available, please also include ORCiDs and social media handles (Facebook, Twitter or LinkedIn). One author will need to be identified as the corresponding author, with their email address normally displayed in the article PDF (depending on the journal) and the online article. Authors' affiliations are the affiliations where the research was conducted. If any of the named co-authors moves affiliation during the peer-review process, the new affiliation can be given as a footnote. Please note that no changes to affiliation can be made after your paper is accepted. Read more on authorship.
2. Should contain a structured abstract of 250 words. The structured abstract should contain the following subheadings: Background; Method; Results; Conclusions. The abstract should

be included in both the main document and uploaded separately. Do not use proprietary names in the abstract.

3. **Graphical abstract** (mandatory). This is an image to give readers a clear idea of the content of your article. It should be a maximum width of 525 pixels. If your image is narrower than 525 pixels, please place it on a white background 525 pixels wide to ensure the dimensions are maintained. Save the graphical abstract as a .jpg, .png, or .tiff. Please do not embed it in the manuscript file but save it as a separate file, labelled GraphicalAbstract1.
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5. Between 3 and 7 **keywords**. Read making your article more discoverable, including information on choosing a title and search engine optimization.
6. **Funding details**. Please supply all details required by your funding and grant-awarding bodies as follows:
For single agency grants
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This work was supported by the [Funding Agency #1] under Grant [number xxxx]; [Funding Agency #2] under Grant [number xxxx]; and [Funding Agency #3] under Grant [number xxxx].
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8. **Biographical note**. Please supply a short biographical note for each author. This could be adapted from your departmental website or academic networking profile and should be relatively brief (e.g. no more than 200 words).
9. **Data availability statement**. If there is a data set associated with the paper, please provide information about where the data supporting the results or analyses presented in the paper can be found. Where applicable, this should include the hyperlink, DOI or other persistent identifier associated with the data set(s). Templates are also available to support authors.
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14. **Tables**. Tables should present new information rather than duplicating what is in the text. Readers should be able to interpret the table without reference to the text. Please supply editable files.
15. **Equations**. If you are submitting your manuscript as a Word document, please ensure that equations are editable. More information about mathematical symbols and equations.
16. **Units**. Please use SI units (non-italicized).

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Disclosure Statement

Please include a disclosure statement, using the subheading “Disclosure of interest.” If you have no interests to declare, please state this (suggested wording: *The authors report no conflict of interest*). For all NIH/Wellcome-funded papers, the grant number(s) must be included in the declaration of interest statement. Read more on declaring conflicts of interest.

Clinical Trials Registry

In order to be published in a Taylor & Francis journal, all clinical trials must have been registered in a public repository at the beginning of the research process (prior to patient enrolment). Trial registration numbers should be included in the abstract, with full details in the methods section. The registry should be publicly accessible (at no charge), open to all prospective registrants, and managed by a not-for-profit organization. For a list of registries that meet these requirements, please visit the WHO International Clinical Trials Registry Platform (ICTRP). The registration of all clinical trials facilitates the sharing of information among clinicians, researchers, and patients, enhances public confidence in research, and is in accordance with the ICMJE guidelines.

Complying With Ethics of Experimentation

Please ensure that all research reported in submitted papers has been conducted in an ethical and responsible manner, and is in full compliance with all relevant codes of experimentation and legislation. All papers which report in vivo experiments or clinical trials on humans or animals must include a written statement in the Methods section. This should explain that all work was conducted with the formal approval of the local human subject or animal care committees (institutional and national), and that clinical trials have been registered as legislation requires. Authors who do not have formal ethics review committees should include a statement that their study follows the principles of the Declaration of Helsinki.

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user, or participant (or that person's parent or legal guardian) in any research, experiment, or clinical trial described in your paper has given written consent to the inclusion of material pertaining to themselves, that they acknowledge that they cannot be identified via the paper; and that you have fully anonymized them. Where someone is deceased, please ensure you have written consent from the family or estate. Authors may use this Patient Consent Form, which should be completed, saved, and sent to the journal if requested.

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Authors are further encouraged to cite any data sets referenced in the article and provide a Data Availability Statement.

At the point of submission, you will be asked if there is a data set associated with the paper. If you reply yes, you will be asked to provide the DOI, pre-registered DOI, hyperlink, or other persistent identifier associated with the data set(s). If you have selected to provide a pre-registered DOI, please be prepared to share the reviewer URL associated with your data deposit, upon request by reviewers.

Where one or multiple data sets are associated with a manuscript, these are not formally peer reviewed as a part of the journal submission process. It is the author's responsibility to ensure the soundness of data. Any errors in the data rest solely with the producers of the data set(s).

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Updated 5-02-2019

Appendix B – Systematic Review Protocol

Systematic Review Protocol

A systematic review of interventions to support the communication of bad news in paediatric settings.

Review question

What evidence exists for the use of interventions to improve/support communication of bad news by clinicians in paediatric and children services?

Searches

Searches of biomedical, psychological, allied health electronic bibliographic databases will be conducted to identify relevant articles. These are: Ovid (PsychINFO, Embase, MEDLINE), EBSCOhost (CINAHL, ERIC) and ProQuest (ASSIA, Dissertations and Theses Global). Additional hand searching will consider relevant articles from the reference lists of included articles. Grey literature will be searched using ProQuest (Dissertations and Theses Global). Non-English language papers will be excluded.

Search terms:

1. Bad/difficult/negative news/conversation
2. Deliver*/inform*/communicat*
3. Paediatric* OR pediatric*/child*

Types of study to be included

Quantitative interventions designs will be included in this review. These will include any identified randomised control trials (RCTs), non-randomised control trials (nRCTs), controlled before and after studies (CBAs) and Interrupted Time Series (ITS) as identified by Cochrane handbook. Less robust designs will also be included for interventions not using designs listed above (though a pre- and post-measure must be included).

Condition or domain being studied

The breaking of “bad news” in paediatrics services. This could be the delivery of difficult or negative information, for example, the diagnosis of a health condition.

Participants/population

Qualified or training clinical staff working in paediatric or children’s services. Any paediatric or child service will be included.

Intervention(s), exposure(s)

This review will include all interventions aimed at improving clinicians' delivery of "bad news". All intervention formats will be included, such as educational, skills training, specific protocols at both person and organisational/systemic levels.

Comparator(s)/control

Any form of control or studies with and without controls will be reviewed. Those without will need to include pre/post measures.

Context

Studies of clinicians (qualified and trainees) working in paediatric and/or children's services will be included for review.

Main outcome(s)

Ability or skill in delivering "bad news" as rated by an observer. This may be any observer, for example, staff member, patient or simulated patient.

Data extraction (selection and coding)

- Retrieved literature will be reviewed and duplicates removed
- Titles and abstracts of the search outcomes will be screened by first reviewer using inclusion/exclusion criteria.
- Full text articles will be obtained for all articles meeting inclusion criteria following initial screening.
- Full text articles will be reviewed and those continuing to meet inclusion criteria identified.
- First reviewer extracts data - Study (design), Participants, Intervention (provide description), Outcomes measured, Results/Analysis, Author conclusions

Risk of bias (quality) assessment

Quality assessment will be completed using Downs and Black checklist (1998). At least 50% papers will be rated independently by two raters.

Appendix C – Final Consensus Quality Assessment Ratings for each criterion for each study

No.	First author and year	REPORTING																			
		1. Is the hypothesis/aim/objective of the study clearly described?		2. Are the main outcomes to be measured clearly described in the Introduction or Methods section?		3. Are the characteristics of the patients included in the study clearly described ?		4. Are the interventions of interest clearly described?		5. Are the distributions of principal confounders in each group of subjects to be compared clearly described?		6. Are the main findings of the study clearly described?		7. Does the study provide estimates of the random variability in the data for the main outcomes?		8. Have all important adverse events that may be a consequence of the intervention been reported?		9. Have the characteristics of patients lost to follow-up been described?		10. Have actual probability values been reported (e.g. 0.035 rather than <0.05) for the main outcomes except where the probability value is less than 0.001?	
		Yes (= 1)	No (= 0)	Yes (= 1)	No (= 0)	Yes (= 1)	No (= 0)	Yes (= 1)	No (= 0)	Yes (= 1)	No (= 0)	Yes (= 1)	No (= 0)	Yes (= 1)	No (= 0)	Yes (= 1)	No (= 0)	Yes (= 1)	No (= 0)	Yes (= 1)	No (= 0)
1	Attar 2010	1		1			0	1			0	1		1			0	1		1	
2	Calhoun 2017	1		1		1		1		1		1		1		1		1		1	
3	Cannone 2019		0	1			0	1			0	1		1			0	1		1	
4	Greenberg 1999	1		1		1		1		1		1		1			0	1		1	
5	Nellis 2014	1		1			0	1			0	1			0		0	1		1	
6	Reed 2015	1		1			0	1			0	1		1			0	1		1	
7	Silva 2018		0	1			0	1			0		0		0		0	1		1	
8	Tobler 2014	1		1			0	1			0	1		1		1			0	1	
9	Vaidya 1999	1		1		1		1		1		1			0		0	1		1	
10	Yuan 2019	1		1		1		1		1		1		1			0	1		1	

No.	First author and year	EXTERNAL VALIDITY									INTERNAL VALIDITY - BIAS														
		11. Were the subjects asked to participate in the study representative of the entire population from which they were recruited?			12. Were those subjects who were prepared to participate representative of the entire population from which they were recruited?			13. Were the staff, places, and facilities where the patients were treated, representative of the treatment the majority of patients receive?			14. Was an attempt made to blind study subjects to the intervention they have received ?			15. Was an attempt made to blind those measuring the main outcomes of the intervention?			16. If any of the results of the study were based on "data dredging", was this made clear?			17. In trials and cohort studies, do the analyses adjust for different lengths of follow-up of patients, or in case-control studies, is the time period between the intervention and outcome the same for cases and controls ?			18. Were the statistical tests used to assess the main outcomes appropriate?		
		Yes (1)	No (0)	Unable to determine	Yes (1)	No (0)	Unable to determine	Yes (1)	No (0)	Unable to determine	Yes (1)	No (0)	Unable to determine	Yes (1)	No (0)	Unable to determine	Yes (1)	No (0)	Unable to determine	Yes (1)	No (0)	Unable to determine	Yes (1)	No (0)	Unable to determine
1	Attar 2010	1					0	1				0			0		1			1			1		
2	Calhoun 2017	1					0	1				0			0		1			1			1		
3	Cannone 2019			0			0	1				0		1			1			1			1		
4	Greenberg 1999			0			0	1				0		1			1				0		1		
5	Nellis 2014			0			0	1				0		1			1			1			1		
6	Reed 2015			0	1			1				0			0		1			1			1		
7	Silva 2018			0			0	1				0		0				0		1			1		
8	Tobler 2014			0			0	1				0		1			1			1			1		
9	Vaidya 1999	1			1			1				0		1			1			1			1		
10	Yuan 2019			0			0	1				0		1			1				0		1		

No.	First author and year	INTERNAL VALIDITY - BIAS						INTERNAL VALIDITY - CONFOUNDING (SELECTION BIAS)								
		19. Was compliance with the intervention/s reliable?			20. Were the main outcome measures used accurate (valid and reliable)?			21. Were the patients in different intervention groups (trials and cohort studies) or were the cases and controls (case-control studies) recruited from the same population?			22. Were study subjects in different intervention groups (trials and cohort studies) or were the cases and controls (case-control studies) recruited over the same period of time?			23. Were study subjects randomised to intervention groups?		
		Yes (= 1)	No (= 0)	Unable to determine	Yes (= 1)	No (= 0)	Unable to determine	Yes (= 1)	No (= 0)	Unable to determine	Yes (= 1)	No (= 0)	Unable to determine	Yes (= 1)	No (= 0)	Unable to determine
1	Attar 2010	1			1				0		1				0	
2	Calhoun 2017	1			1			1			1			1		
3	Cannone 2019			0	1				0			0			0	
4	Greenberg 1999	1			1			1			1				0	
5	Nellis 2014	1			1			1			1				0	
6	Reed 2015	1			1			1			1				0	
7	Silva 2018	1			1			1			1				0	
8	Tobler 2014	1					0	1					0		0	
9	Vaidya 1999	1			1			1			1			1		
10	Yuan 2019	1			1			1			1			1		

INTERNAL VALIDITY - CONFOUNDING (SELECTION BIAS)											POWER					
No.	First author and year	24. Was the randomised intervention assignment concealed from both patients and health care staff until recruitment was complete and irrevocable?			25. Was there adequate adjustment for confounding in the analyses from which the main findings were drawn?			26. Were losses of patients to follow-up taken into account?			27. Did the study have sufficient power to detect a clinically important effect where the probability value for a difference being due to chance is less than 5%?					
		Yes (= 1)	No (= 0)	Unable to determine (=0)	Yes (= 1)	No (= 0)	Unable to determine (=0)	Yes (= 1)	No (= 0)	Unable to determine (=0)	Size of smallest intervention group					
											<n1 (=0)	n1–n2 (=1)	n3–n4 (=2)	n5–n6 (=3)	n7–n8 (=4)	n8+ (=5)
1	Attar 2010		0				0	1								5
2	Calhoun 2017		0		1			1								5
3	Cannone 2019		0			0		1								5
4	Greenberg 1999		0		1			1								5
5	Nellis 2014		0				0	1								5
6	Reed 2015		0				0	1								5
7	Silva 2018		0				0	1						3		5
8	Tobler 2014		0				0			0						5
9	Vaidya 1999			0		0		1							4	
10	Yuan 2019			0	1			1								5

Appendix D: Author Guidelines for Journal of Genetic Counselling

1. AIMS AND SCOPE

The **Journal of Genetic Counseling (JOGC)**, published for the National Society of Genetic Counselors, is a timely, international forum addressing all aspects of the discipline and practice of genetic counseling. The journal focuses on the critical questions and problems that arise at the interface between rapidly advancing technological developments and the concerns of individuals and communities at genetic risk. The publication provides genetic counselors, other clinicians and health educators, laboratory geneticists, bioethicists, legal scholars, social scientists, and other researchers with a premier resource on genetic counseling topics in national, international, and cross-national contexts.

As a crucial resource for genetic counselors and associated professionals, the Journal's primary purpose is to report original research in the following areas:

- **Genetic Counseling Theory, Methods, and Practice:** addresses genetic counseling in clinical or non-clinical settings;
- **Public Health, Public Policy, and Access and Genetics Service Delivery:** addresses public health genomics, health behaviors, legal or policy aspects related to genetic counseling and genetic testing, precision medicine, health disparities, models of genetics services delivery;
- **Education and Genetics Professional Workforce Issues:** addresses educational training, professional development, and workforce topics related to genetic counseling;
- **Ethical, Legal, Psychological, and Social Issues:** addresses ethical, legal, psychological, and/or social issues related to genetic counseling, genetic services, and/or genetic information regarding individuals, communities, and the public
- **Risk Assessment:** addresses algorithms, theoretical models, or empirical data for use in genetic counseling risk assessment.

In addition to research articles, regular features of the Journal of Genetic Counseling include case presentations, editorials, rapid publications, and letters to the editor. Note: The Journal does not publish non-human animal studies.

2. SUBMISSION

Once the submission materials have been prepared in accordance with the Author Guidelines, manuscripts should be submitted via the journal's Editorial Manager site: <https://www.editorialmanager.com/jogc/default.aspx>. More details on how to use Editorial Manager are also available at <https://www.editorialmanager.com/jogc/default.aspx>.

A manuscript is considered for review and possible publication on the condition that it is submitted solely to the journal, and that the manuscript or a substantial portion of it is not under consideration elsewhere. Presentation of the content at meetings prior to submission is acceptable. However, authors should kindly note that submission implies that the content has not been published or submitted for publication elsewhere *except* as a brief abstract in the proceedings of a scientific meeting or symposium. Note, this journal uses iThenticate's CrossCheck software to detect instances of overlapping and similar text in submitted manuscripts.

The submission system will prompt the author to use an ORCID ID (a unique author identifier) to help distinguish their work from that of other researchers. [Click here](#) to find out more.

For help with submissions, please contact the Editorial Office at JOGC@Wiley.com. When necessary, the Editorial Office staff may refer questions to the Editor-in-Chief.

3. MANUSCRIPT CATEGORIES AND GENERAL REQUIREMENTS

MANUSCRIPT CATEGORIES

Original Articles. The Journal of Genetic Counseling seeks papers reporting exciting, timely, original research in the discipline and practice of genetic counseling. The Journal considers papers using a form of systematic study or inquiry to address a question to be original research. Systematic study can be approached using a variety of methods, such as empirical methods, systematic literature review methods, normative or conceptual research methods. Original articles:

- include an abstract and key words;
- are no more than 25 double-spaced pages in length for quantitative studies and no more than 35 double-spaced pages in length for qualitative or non-empirical studies (excluding Supplemental Information);
- have no more than 5 display items (tables + figures), and any additional display items will need to be submitted as Supplemental Information. Large tables should always be published as online only material;
- report relevant information per appropriate methodologic guideline (see Research Reporting Guidelines below).

Case Studies. Case studies are a valuable tool in the presentation of genetic counseling practice. They can serve to demonstrate a counseling model or to stimulate thought about a difficult ethical or counseling situation the author has encountered. In a case study, the paper is focused on the case(s) presented with the intention of alerting the reader to broader issues relevant to practice for the readers' consideration. Note: the Journal of Genetic Counseling does not publish case studies whose sole purpose is to report clinical and molecular information. Case Studies should be concise and focused. They should address observations of patient encounters (usually 1-3) or a single small family that add substantially to the practice and discipline of genetic counseling. Case Studies:

- include an abstract and key words;
- are no more than 15 double spaced pages in length (excluding Supplemental Information);
- have no more than 2 display items (tables + figures), and any additional display items will need to be submitted as Supplemental Information. Large tables should always be published as online only material.

Professional Issues. These article types feature pieces that communicate reflections by the author on the discipline and practice of genetic counseling. Professional Issues:

- include an abstract and key words;
- are no more than 25 double-spaced pages in length (excluding Supplemental Information);
- have no more than 2 display items (tables + figures). and any additional display items will need to be submitted as Supplemental Information. Large tables should always be published as online only material.

Invited Commentary. This type of paper is generally solicited from the Editor but is a submission welcomed from all contributors. It should have a title page and be accompanied by a list of key words for indexing purposes. Commentaries/Editorials often address matters of interest or controversy to the readership.

Brief Reports. These are very brief reports offered in a letter format reporting an observation that adds to the knowledge of the discipline and practice of genetic counseling. They are no more than 9 double spaced manuscript pages in length (excluding Supplemental Information). The

manuscripts are not subdivided into sections nor do they include an abstract. Key words are required for indexing purposes.

Correspondence. These are letters to the editor and generally comment on previously published work in the Journal of Genetic Counseling. These are kept brief and to the point; they do not include an abstract, key words, tables, or figures. Like all other material published in the Journal of Genetic Counseling, correspondence is subject to editorial or peer review. The corresponding author of the original manuscript which is the subject of the submitted letter will be offered the opportunity to respond. If a response is provided, every effort will be made to publish these letters together. Only one round of comment is allowed.

Rapid Communications. The Journal of Genetic Counseling features a new section devoted to the rapid communication of full-length, critically reviewed papers reporting new and important advances that are highly likely to have an immediate and critical impact on the discipline and practice of genetic counseling. Our goal is that these manuscripts will be published online approx. 4 weeks after acceptance. In order to have a manuscript considered for Rapid Communication, authors must send a letter of intent along with an abstract to the Editor for consideration prior to submission. The letter of intent should outline the author's rationale for publishing the article as a rapid publication. The Editor or Deputy Editor will respond to the author with a decision. Manuscripts accepted for Rapid publication must adhere to the format of an original research article in the Journal of Genetic Counseling.

Practice Guidelines. These article types address specific areas of genetic counseling and are submitted by the National Society of Genetic Counselors' Practice Guidelines Committee.

Review Articles. The Journal of Genetic Counseling publishes occasional topical reviews. Authors should contact the Editor-in-Chief prior to submission. Note: submissions that describe a systematic process for reviewing the literature to address a research question (e.g., systematic reviews, scoping reviews) are considered original research and are included in the Original Article category.

Book Reviews. Authors may contact the Editor-in-Chief with a proposal to submit a book review. The topic of the reviewed book should be closely aligned with the mission of the Journal. If the proposal is approved for the submission, instruction will be provided by the editor.

Conference Reports. The Journal of Genetic Counseling occasionally publishes an executive summary of an important conference or scientific meeting that involves topics related to the scope of the Journal. The Journal also on occasion publishes the abstracts of an important meeting on a selected basis. Authors should contact the Editor-in-Chief prior to submission.

Corrigenda and Errata. These manuscripts are brief communications to correct errors in previously published work in the Journal of Genetic Counseling. The former is for errors that were responsibility of the author(s), and the latter are for errors that are responsibility of the Journal, including editorial staff and production. These may be written by the corresponding author of the relevant manuscript or they may be composed by an editor.

GENERAL REQUIREMENTS

Format

Manuscripts should be double-spaced with 1 inch margins and 12 point font.

English Language

Manuscripts must be submitted in grammatically correct American English. Manuscripts that do not meet this standard cannot be reviewed. Authors for whom English is a second language may wish to consult an English-speaking colleague or consider having their manuscript professionally edited before submission to improve the English. A list of independent suppliers of editing services can be found at <https://wileyeditingservices.com/en/>. All services are paid for and arranged by the author, and use of one of these services does not guarantee acceptance or preference for publication.

Revisions

Please submit a marked version (tracked, highlighted, etc) and unmarked version of revised manuscripts.

Ethical Compliance

For all research involving human participants, please include a statement in the Methods section confirming that your study was reviewed by an institutional review board/human investigations committee/ethics committee (include name of committee) and approved or waived as human subjects research.

The Journal of Genetic Counseling does not publish research involving non-human animals.

Informed Consent

The Journal requires that all appropriate steps be taken in obtaining informed consent of all human subjects participating in the research comprising the manuscript submitted for review and possible publication, and statements to this effect must be included under the subheadings, "Human Studies and Informed Consent". For all manuscript categories, identifying information should not be included in the manuscript unless the information is essential for scientific purposes and the study participants or patients (or parents or guardians) give written informed consent for publication. The editors reserve the right to reject manuscripts that do not comply with these requirements. The author will be held responsible for false statements or failure to fulfill these requirements.

Conflict of Interest Statement

The Journal requires that all authors disclose any potential sources of conflict of interest. Any interest or relationship, financial or otherwise, that might be perceived as influencing an author's objectivity is considered a potential source of conflict of interest. These must be disclosed when directly relevant or directly related to the work that the authors describe in their manuscript. Potential sources of conflict of interest include, but are not limited to, patent or stock ownership, membership of a company board of directors, membership of an advisory board or committee for a company, and consultancy for or receipt of speaker's fees from a company. The existence of a conflict of interest does not preclude publication in this journal.

If the authors have no conflict of interest to declare, they must also state this in the manuscript. It is the responsibility of the corresponding author to review this policy with all authors and collectively to list in the manuscript under the subheading "Conflict of Interest" ALL pertinent commercial and other relationships.

The above policies are in accordance with the Uniform Requirements for Manuscripts Submitted to Biomedical Journals produced by the International Committee of Medical Journal Editors (<http://www.icmje.org/>).

4. PREPARING THE SUBMISSION

Parts of the Manuscript

The manuscript should be submitted in separate files: cover letter; main text file; tables; figures; supplementary information files.

Cover Letter

The cover letter should include a statement that the work presented in the manuscript has not been published elsewhere and is not currently under review elsewhere.

If the study includes original data, at least one author must confirm in the cover letter that he or she had full access to all the data in the study and takes responsibility for the integrity of the data and the accuracy of the data analysis.

Main Text File

The main text file should be presented in the following order (as appropriate for article type):

1. Title Page
2. Abstract and keywords
3. Main text
4. Author Contributions

5. Acknowledgements
6. Conflict of Interest
7. Human Studies and Informed Consent
8. Animal Studies
9. References
10. Figure legends

Tables, figures and supplementary information files should be supplied as separate files. Figures must be clearly labeled.

Title Page

The title page should include (in this order) the title of the article, authors' names (no degrees), authors' institutional affiliations where the work was conducted, with a footnote for the author's present address if different from where the work was conducted, and suggested running head. The affiliation should comprise the department, institution (usually university or company), city, and state (or nation) and should be typed as a numbered footnote to the author's name. The suggested running head should be less than 80 characters (including spaces) and should comprise the article title or an abbreviated version thereof. The title page should also include the telephone number and e-mail address of the one author designated to review proofs.

Please denote cases of equal authorship with a footnote: In the case of joint first authorship, a footnote should be added to the author listing, e.g. 'X and Y should be considered joint first author' or 'X and Y should be considered joint senior author'.

Authors may benefit from referring to Wiley's best practice tips on [Writing for Search Engine Optimization](#).

Abstract

Please provide an unstructured abstract of no more than 300 words containing the major keywords summarizing the article. The abstract should include a description of the study's objective, methods or methodological approach, sample, measures or main outcome variables, main results, and conclusion.

Keywords

Please provide three to six keywords to be used for indexing the article. Please refer to [this list](#).

Main Body

For Original Research articles, all major sections should carry section headings (such as Introduction, Methods, Results, Discussion, Conclusions, etc.) type centered. Side headings in Methods section should include, as appropriate: Participants, Instrumentation, Procedures, and Data Analysis. The Discussion should begin with a very succinct summary of the major conclusions of the paper and then go on to focus on the interpretation and significance of the findings with concise objective comments that describe their relation to other work in the area. It should not repeat information in the results. Side headings in Discussion should include: Study Limitations, Practice Implications, and Research Recommendations. The journal uses US spelling.

Footnotes should be avoided in the main text. When their use is absolutely necessary, footnotes should be numbered consecutively using Arabic numerals and should be typed at the bottom of the page to which they refer. Place a line above the footnote, so it is set off from the text. Use the appropriate superscript numeral for citation in the text.

Author Contributions

Please include a statement delineating the contributions of each author using the criteria recommended by the International Committee of Medical Journal Editors (ICMJE). The statement should mention each author separately by name. ICMJE criteria are:

- Substantial contributions to the conception or design of the work; or the acquisition, analysis, or interpretation of data for the work; AND
- Drafting the work or revising it critically for important intellectual content; AND

- Final approval of the version to be published; AND
- Agreement to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

If the study includes original data, at least one author must confirm that he or she had full access to all the data in the study and takes responsibility for the integrity of the data and the accuracy of the data analysis. Please include this statement in the cover letter.

Acknowledgements

Contributions from anyone who does not meet the criteria for authorship should be listed, with permission from the contributor, in an Acknowledgments section. Financial and material support should also be mentioned. Authors should list all funding sources and are responsible for the accuracy of their funder designation. If in doubt, please check the Open Funder Registry for the correct nomenclature: www.crossref.org/services/funder-registry.

If this paper is to be considered for the Journal of Genetic Counseling Best Trainee Paper award, please include a statement indicating that the research presented in the paper was conducted while the first author was in training or to fulfill a degree requirement of the first author. See the [Best Trainee Paper Award](#) tab on the journal website for more information about this award. Thanks to anonymous reviewers is not considered appropriate to include in Acknowledgements.

Conflict of Interest Statement

The Conflict of Interest Statement should mention each author separately by name. Recommended wording is as follows:

Author X declares that she has no conflict of interest.

Author Y has received research grants from Drug Company A.

Author Z has received a speaker honorarium from Drug Company B and owns stock in Drug Company C.

If multiple authors declare no conflict, this can be done in one sentence:

Author X, Author Y and Author Z declare that they have no conflict of interest.

Submitting authors should ensure they liaise with all co-authors to confirm agreement with the final statement.

Human Studies and Informed Consent

For manuscripts reporting studies that involve human participants, a statement identifying the ethics committee that approved the study and confirmation that the study conforms to recognized standards is required, for example: [Declaration of Helsinki](#); [US Federal Policy for the Protection of Human Subjects](#); or [European Medicines Agency Guidelines for Good Clinical Practice](#). It should also state clearly in the text that all persons gave their informed consent prior to their inclusion in the study.

The Journal requires that all appropriate steps be taken in obtaining informed consent of any and all human subjects participating in the research comprising the manuscript submitted for review and possible publication, and a statement to this effect must be included in the Human Studies and Informed Consent section of the manuscript. Participant anonymity should be preserved and all identifying information should be excluded in the manuscript.

Photographs need to be cropped sufficiently to prevent human subjects being recognized (an eye bar must not be used because of insufficient de-identification). Images and information from individual participants will only be published where the authors have obtained the individual's free prior informed consent. If any identifying information about participants is included in the article, the following sentence should also be included:

'Additional informed consent was obtained from all participants for which identifying information is included in this article.'

Authors do not need to provide a copy of the consent form to the publisher; however, in signing the author license to publish, authors are required to confirm that consent has been obtained.

Wiley has a [standard patient consent form available](#) for use.

Animal Studies

The Journal of Genetic Counseling does not publish non-human animal studies. To affirm that this is the case for your submission, please include the following sentence under this subheading in the manuscript:

'No non-human animal studies were carried out by the authors for this article'

References

The accuracy of references is the responsibility of the authors. Only published papers and those in press may be included in the reference list. The Journal has a strong preference against the inclusion of conference abstracts (published or unpublished) or unpublished data in manuscripts. However, if done, unpublished data and submitted manuscripts must be cited parenthetically within the text. Personal communications should also be cited within the text; permission in writing from the communicator is required.

References should be prepared according to the *Publication Manual of the American Psychological Association* (6th edition). The APA website includes a range of [resources for authors](#) learning to write in APA style, including [an overview](#) of the manual, [free tutorials](#) on APA Style basics, and an [APA Style Blog](#). For more information about APA referencing style, please also refer to the [APA FAQ](#).

EndNote users can download the style [here](#).

According to APA style, in text citations should follow the author-date method whereby the author's last name and the year of publication for the source should appear in the text, for example, (Jones, 1998). Multiple citations should be listed alphabetically by author's last name. The complete reference list should appear alphabetically by name at the end of the paper.

Authors should note that the APA referencing style requires that a Digital Object Identifier (DOI) be provided for all references where available. Also, for journal articles, issue numbers are not included unless each issue in the volume begins with page one.

Reference examples follow:

Journal article with fewer than 7 authors

Beers, S. R., & De Bellis, M. D. (2002). Neuropsychological function in children with maltreatment-related posttraumatic stress disorder. *The American Journal of Psychiatry*, 159(2), 483–486. doi:10.1176/appi.ajp.159.3.483

Journal article with 7 or more authors

Shelton, B. A., John, D., Gibbs, J. T., Huang, L. N., Ruble, D. N., Martin, C. L., ... Seltzer, M. M. (1996). The division of household labor. *Annual Review of Sociology*, 22, 299–322. doi:10.1146/annurev.soc.22.1.299

Note: for more than seven author names list first six with three dots and then last author name.

Book

Bradley-Johnson, S. (1994). *Psychoeducational assessment of students who are visually impaired or blind: Infancy through high school* (2nd ed.). Austin, TX: Pro-ed.

Internet Document

Norton, R. (2006, November 4). How to train a cat to operate a light switch [Video file]. Retrieved from <http://www.youtube.com/watch?v=Vja83KLQXZs>

Figure Legends

Every figure must have a legend. Legends should be concise but comprehensive – the figure and its legend must be understandable without reference to the text. Include definitions of any symbols used and define/explain all abbreviations and units of measurement. Figures should be numbered (with Arabic numerals) and referred to by number in the text.

Additional Files

Tables

Tables should be self-contained and complement, not duplicate, information contained in the text. Tables should be numbered (with Arabic numerals) and referred to by number in the text. They should be supplied as editable files, not pasted as images. The table should have a brief explanatory title, and legends should be concise but comprehensive – the table, legend, and footnotes must be understandable without reference to the text. All abbreviations must be defined in table footnotes. Footnotes should be indicated by superscript lowercase letters and *, **, *** should be reserved for P-values. Statistical measures such as SD or SEM should be identified in the table headings. Each table should be on a separate sheet of paper at the end of the submission.

Figures

Authors are encouraged to send the highest quality figures possible. Line art should be exported at 600 dpi or higher, and halftone images should be exported at 300 dpi or higher.

Supporting Information

Supporting information is information that is not essential to the article, but provides greater depth and background. It is hosted online and appears without editing or typesetting. It may include copies of surveys or interview questions, consent forms, tables, figures, videos, datasets, etc.

[Click here](#) for Wiley's FAQs on Supporting Information.

Note: if data, scripts, or other artefacts used to generate the analyses presented in the paper are available via a publicly available data repository, authors should include a reference to the location of the material within their paper.

General Style Points

The following points provide general advice on formatting and style.

- **Abbreviations:** In general, terms should not be abbreviated unless they are used repeatedly and the abbreviation is helpful to the reader. Initially, use the word in full, followed by the abbreviation in parentheses. Thereafter use the abbreviation only.
- **Units of measurement:** Measurements should be given in SI or SI-derived units. Visit the [Bureau International des Poids et Mesures \(BIPM\) website](#) for more information about SI units.
- **Numbers:** numbers under 10 should be spelled out, except for: measurements with a unit (8 mmol/L); age (6 weeks old), or lists with other numbers (11 dogs, 9 cats, 4 gerbils).
- **Trade Names:** Chemical substances should be referred to by the generic name only. Trade names should not be used. Drugs should be referred to by their generic names. If proprietary drugs have been used in the study, refer to these by their generic name, mentioning the proprietary name and the name and location of the manufacturer in parentheses.
- **Genomic Terminology and Nomenclature:** Please use the following terms: genome sequencing instead of whole genome sequencing; exome sequencing instead of whole exome sequencing; pathogenic variant instead of mutation; secondary finding instead of incidental finding. Please italicize gene names; do not italicize protein names. Sequence variants should be described in the text and tables using both DNA and protein designations whenever appropriate. Sequence variant nomenclature must follow the current HGVS guidelines; see varnomen.hgvs.org, where examples of acceptable nomenclature are provided. Human gene nomenclature should follow the standards of the HUGO Gene Nomenclature Committee (HGNC), see <https://www.genenames.org/>.
- **Pedigrees:** Pedigrees should follow the recommendations for standardized nomenclature accepted by the National Society of Genetic Counselors. Authors should consult the following references for these recommendations:
 - Bennett, R. L. , Steinhaus, K. A., Uhrich, S. B., O' Sullivan, C. K., Resta, R. G. , Lochner-Doyle, D., Markel, D. S., Vincent, V., & Hamanishi, J. (1995).

Recommendations for Standardized Human Pedigree Nomenclature. *Journal of Genetic Counseling*, 4, 267-279.

- Bennett, R. L., Steinhaus French, K., Resta, R. G., & Lochner Doyle, D. (2008). Standardized Human Pedigree Nomenclature: Update and Assessment of the Recommendations of the National Society of Genetic Counselors. *Journal of Genetic Counseling*, 17, 424-433.

5. EDITORIAL POLICIES AND ETHICAL CONSIDERATIONS

Peer Review and Acceptance

The acceptance criteria for all papers are the quality and originality of the research and its significance to journal readership and the practice and discipline of genetic counseling. Papers will only be sent to review if the Editors determine that the paper meets the appropriate quality and relevance requirements.

Except where otherwise stated, manuscripts are single-blind peer reviewed. Wiley's policy on the confidentiality of the review process is [available here](#).

Data Sharing and Data Accessibility

The *Journal of Genetic Counseling* expects that data supporting the results in the paper will be archived in an appropriate public repository. Authors are required to provide a data availability statement to describe the availability or the absence of shared data. When data have been shared, authors are required to include in their data availability statement a link to the repository they have used, and to cite the data they have shared. Whenever possible the scripts and other artefacts used to generate the analyses presented in the paper should also be publicly archived. If sharing data compromises ethical standards or legal requirements then authors are not expected to share it.

Although it would be rare for a paper submitted to the *Journal of Genetic Counseling* to report novel nucleotide sequence data, should that be the case, the novel nucleotide sequence data including genetic mutations must be submitted to a public database prior to publication and a sentence naming the database should be included in the manuscript.

Human Studies and Subjects

For manuscripts reporting studies that involve human participants, a statement identifying the institutional review board/human investigations committee/ethics committee that approved the study and confirmation that the study conforms to recognized standards is required. It should also state clearly in the text that all persons gave their informed consent prior to their inclusion in the study.

Patient anonymity should be preserved. Information from individual patients will only be published where the authors have obtained the individual's free prior informed consent. Authors do not need to provide a copy of the consent form to the publisher; however, in signing the author license to publish, authors are required to confirm that consent has been obtained. Wiley has a [standard patient consent form available](#) for use if needed.

Clinical Trial Registration

The *Journal* requires that clinical trials are prospectively registered in a publicly accessible database and clinical trial registration numbers are included in all papers that report their results. Authors are asked to include the name of the trial register and the clinical trial registration number at the end of the Abstract. If the trial is not registered, or was registered retrospectively, the reasons for this should be explained.

Research Reporting Guidelines

Accurate and complete reporting enables readers to fully appraise research, replicate it, and use it. Authors are encouraged to adhere to recognized research reporting standards. The EQUATOR Network collects more than 370 reporting guidelines for many study types, including for:

- [Randomized trials: CONSORT](#)
- [Observational studies: STROBE](#)
- [Systematic reviews: PRISMA](#)
- [Qualitative research: COREQ](#)
- [Quality improvement studies: SQUIRE](#)
- [Study protocols: SPIRIT](#)

Studies reporting on genetic counseling as an intervention should refer to and follow guidelines from:

- Standards for the Reporting of Genetic Counseling Interventions in Research and Other Studies (GCIRS), available [here](#).

Publication Ethics

This journal is a member of the [Committee on Publication Ethics \(COPE\)](#). Read Wiley's Top 10 Publishing Ethics Tips for Authors [here](#). Wiley's Publication Ethics Guidelines can be found [here](#).

Author Guidelines updated October 2018.

Appendix E: Ethical Approval from South East Scotland REC



Lothian NHS Board

South East Scotland Research
Ethics Committee 02

Waverley Gate
2-4 Waterloo Place
Edinburgh
EH1 3EG
Telephone 0131 536 9000

www.nhslothian.scot.nhs.uk

Date 31 July 2019
Your Ref
Our Ref

Enquiries to : Joyce Clearie
Extension: 35674
Direct Line: 0131 465 5674

31 July 2019

Ms Blathnaid Greene
Trainee Clinical Psychologist
NHS Fife
Lynebank Hospital
Halbeath Road
Dunfermline
KY11 4UW

Dear Ms Greene

Study title:	Parental experiences of receiving a newborn diagnosis of cystic fibrosis.
REC reference:	19/SS/0079
Protocol number:	CAHSS1808/01
IRAS project ID:	249098

Thank you for your letter of 22nd July 2019 and email of 31st July 2019, responding to the Committee's request for further information on the above research and submitting revised documentation.

The further information has been considered on behalf of the Committee by the Chair.

Confirmation of ethical opinion

On behalf of the Committee, I am pleased to confirm a favourable ethical opinion for the above research on the basis described in the application form, protocol and supporting documentation as revised, subject to the conditions specified below.



Headquarters
Waverley Gate
2-4 Waterloo Place
Edinburgh EH1 3EG

Chair Brian G. Houston
Chief Executive Tim Davison
*Lothian NHS Board is the common
name of Lothian Health Board*

Conditions of the favourable opinion

The REC favourable opinion is subject to the following conditions being met prior to the start of the study.

1. *Please amend as discussed Cystic Fibrosis Nurse Specialist Information Sheet to include the same information as that included in the PIS: "Once the interview has been transcribed the recording will be destroyed".*
2. *Consent Form wording to be amended to read "person" safety.*
3. *Amend the PIS wording to include: "Once the researcher has analysed the data, she will contact participants who have given their permission to ask for their feedback on the interpretations of the interview. This is completely voluntary and you are under no obligation to provide feedback even if you have taken part in the interview."*

You should notify the REC once all conditions have been met (except for site approvals from host organisations) and provide copies of any revised documentation with updated version numbers. Revised documents should be submitted to the REC electronically from IRAS. The REC will acknowledge receipt and provide a final list of the approved documentation for the study, which you can make available to host organisations to facilitate their permission for the study. Failure to provide the final versions to the REC may cause delay in obtaining permissions.

Confirmation of Capacity and Capability (in England, Northern Ireland and Wales) or NHS management permission (in Scotland) should be sought from all NHS organisations involved in the study in accordance with NHS research governance arrangements. Each NHS organisation must confirm through the signing of agreements and/or other documents that it has given permission for the research to proceed (except where explicitly specified otherwise).

Guidance on applying for HRA and HCRW Approval (England and Wales)/ NHS permission for research is available in the Integrated Research Application System.

For non-NHS sites, site management permission should be obtained in accordance with the procedures of the relevant host organisation.

Sponsors are not required to notify the Committee of management permissions from host organisations

Registration of Clinical Trials

It is a condition of the REC favourable opinion that all clinical trials are registered on a publicly accessible database. For this purpose, clinical trials are defined as the first four project categories in IRAS project filter question 2. For clinical trials of investigational medicinal products (CTIMPs), other than adult phase I trials, registration is a legal requirement.

Registration should take place as early as possible and within six weeks of recruiting the first research participant at the latest. Failure to register is a breach of these approval conditions, unless a deferral has been agreed by or on behalf of the Research Ethics Committee (see here for more information on requesting a deferral: <https://www.hra.nhs.uk/planning-and-improving-research/research-planning/research-registration-research-project-identifiers/>)

As set out in the UK Policy Framework, research sponsors are responsible for making information about research publicly available before it starts e.g. by registering the research project on a publicly accessible register. Further guidance on registration is available at: <https://www.hra.nhs.uk/planning-and-improving-research/research-planning/transparency-responsibilities/>

You should notify the REC of the registration details. We will audit these as part of the annual progress reporting process.

It is the responsibility of the sponsor to ensure that all the conditions are complied with before the start of the study or its initiation at a particular site (as applicable).

After ethical review: Reporting requirements

The attached document "After ethical review – guidance for researchers" gives detailed guidance on reporting requirements for studies with a favourable opinion, including:

- Notifying substantial amendments
- Adding new sites and investigators
- Notification of serious breaches of the protocol
- Progress and safety reports
- Notifying the end of the study, including early termination of the study
- Final report

The latest guidance on these topics can be found at <https://www.hra.nhs.uk/approvals-amendments/managing-your-approval/>.

Ethical review of research sites

NHS/HSC sites

The favourable opinion applies to all NHS/HSC sites listed in the application subject to confirmation of Capacity and Capability (in England, Northern Ireland and Wales) or management permission (in Scotland) being obtained from the NHS/HSC R&D office prior to the start of the study (see "Conditions of the favourable opinion" below).

Non-NHS/HSC sites if applicable

I am pleased to confirm that the favourable opinion applies to any non-NHS/HSC sites listed in the application, subject to site management permission being obtained prior to the start of the study at the site.

Approved documents

The final list of documents reviewed and approved by the Committee is as follows:

<i>Document</i>	<i>Version</i>	<i>Date</i>
GP/consultant information sheets or letters [08.07.19 Generic GP and Healthcare Professional Letter]	2	08 July 2019
GP/consultant information sheets or letters [08.07.19 NHS Fife GP and Healthcare Professional Letter]	2	08 July 2019
GP/consultant information sheets or letters [08.07.19 NHS Lothian GP and Healthcare Professional Letter]	2	08 July 2019
GP/consultant information sheets or letters [08.07.19 NHS Forth Valley GP and Healthcare Professional Letter]	2	08 July 2019
GP/consultant information sheets or letters [08.07.19 NHS Tayside GP and Healthcare Professional Letter]	2	08 July 2019
Interview schedules or topic guides for participants [26.10.18 Interview Schedule]		26 October 2018
Interview schedules or topic guides for participants [26.10.18 Interview Schedule]	1	26 October 2018
IRAS Application Form [IRAS_Form_08052019]		08 May 2019
IRAS Application Form XML file [IRAS_Form_08052019]		08 May 2019
IRAS Checklist XML [Checklist_29072019]		29 July 2019

Other [Clinical Trial Liability]	1	31 July 2018
Other [Employers' Liability Insurance]	1	01 August 2018
Other [Indemnity Confirmation]	1	24 July 2018
Other [Professional Indemnity Insurance Confirmation]	1	31 July 2018
Other [26.10.18 Academic Supervisor CV Summary]	1	26 October 2018
Other [08.07.19 Generic Cystic Fibrosis Nurse Specialist Information Sheet]	2	08 July 2019
Other [08.07.19 NHS Fife Cystic Fibrosis Nurse Specialist Information Sheet]	2	08 July 2019
Other [08.07.19 NHS Lothian Cystic Fibrosis Nurse Specialist Information Sheet]	2	08 July 2019
Other [08.07.19 NHS Forth Valley Cystic Fibrosis Nurse Specialist Information Sheet]	2	08 July 2019
Other [08.07.19 NHS Tayside Cystic Fibrosis Nurse Specialist Information Sheet]	2	08 July 2019
Other [08.07.19 Generic Demographic Information Sheet]	2	08 July 2019
Other [08.07.19 NHS Fife Demographic Information Sheet]	2	08 July 2019
Other [08.07.19 NHS Lothian Demographic Information Sheet]	2	08 July 2019
Other [08.07.19 NHS Forth Valley Demographic Information Sheet]	2	08 July 2019
Other [08.07.19 NHS Tayside Demographic Information Sheet]	2	08 July 2019
Other [08.07.19 Generic Participant Debriefing Form]	2	08 July 2019
Other [08.07.19 NHS Fife Participant Debriefing Form]	2	08 July 2019
Other [08.07.19 NHS Lothian Participant Debriefing Form]	2	08 July 2019
Other [08.07.19 NHS Forth Valley Participant Debriefing Form]	2	08 July 2019
Other [08.07.19 NHS Tayside Participant Debriefing Form]	2	08 July 2019
Other [Letter for REC with amendment summary table]	1	22 July 2019
Participant consent form [08.07.19 Generic Consent Form]	2	08 July 2019
Participant consent form [08.07.19 NHS Fife Participant Consent Form]	2	08 July 2019
Participant consent form [08.07.19 NHS Lothian Participant Consent Form]	2	08 July 2019
Participant consent form [08.07.19 NHS Forth Valley Participant Consent Form]	2	08 July 2019
Participant consent form [08.07.19 NHS Tayside Participant Consent Form]	2	08 July 2019
Participant information sheet (PIS) [08.07.19 Generic Participant Information Sheet]	2	08 July 2019
Participant information sheet (PIS) [08.07.2019 NHS Fife Participant Information Sheet]	2	08 July 2019
Participant information sheet (PIS) [08.07.2019 NHS Lothian Participant Information Sheet]	2	08 July 2019
Participant information sheet (PIS) [08.07.2019 NHS Forth Valley Participant Information Sheet]	2	08 July 2019
Participant information sheet (PIS) [08.07.19 NHS Tayside Participant Information Sheet]	2	08 July 2019
Research protocol or project proposal [26.10.18 Study Protocol]	1	26 October 2018
Research protocol or project proposal [08.07.19 Study Protocol]	2	08 July 2019
Summary CV for Chief Investigator (CI) [26.10.18 Chief Investigator CV]		26 October 2018

Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

User Feedback

The Health Research Authority is continually striving to provide a high quality service to all applicants and sponsors. You are invited to give your view of the service you have received and the application procedure. If you wish to make your views known please use the feedback form available on the HRA website: <http://www.hra.nhs.uk/about-the-hra/governance/quality-assurance/>

HRA Learning

We are pleased to welcome researchers and research staff to our HRA Learning Events and online learning opportunities– see details at: <https://www.hra.nhs.uk/planning-and-improving-research/learning/>

19/SS/0079

Please quote this number on all correspondence

With the Committee's best wishes for the success of this project.

Yours sincerely

Chair
Mr L Murray

Vice Chair
Prof L Sawyer

Email:

Enclosures: "After ethical review – guidance for researchers"

Copy to: Ms Charlotte Smith

South East Scotland REC 02

2 - 4 Waterloo Place
Edinburgh
EH1 3EG

Telephone: 0131 465 5674
Fax:

28 October 2019

Ms Blathnaid Greene
Trainee Clinical Psychologist
NHS Fife
Lynebank Hospital
Halbeath Road
Dunfermline
KY11 4UW

Dear Ms Greene

Study title: Parental experiences of receiving a newborn diagnosis of cystic fibrosis.
REC reference: 19/SS/0079
Protocol number: CAHSS1808/01
IRAS project ID: 249098

Thank you for uploading additional documents on IRAS on 5th August 2019 in relation to your additional conditions met response. I can confirm the REC has received the documents listed below and that these comply with the approval conditions detailed in our letter dated 31 July 2019

Documents received

The documents received were as follows:

<i>Document</i>	<i>Version</i>	<i>Date</i>
Other [31.07.19 Generic Cystic Fibrosis Nurse Specialist Information Sheet]	3	31 July 2019
Other [31.07.19 NHS Fife Cystic Fibrosis Nurse Specialist Information Sheet]	3	31 July 2019
Other [31.07.19 NHS Lothian Cystic Fibrosis Nurse Specialist Information Sheet]	3	31 July 2019
Other [31.07.19 NHS Forth Valley Cystic Fibrosis Nurse Specialist Information Sheet]	3	31 July 2019
Other [31.07.19 NHS Tayside Cystic Fibrosis Nurse Specialist Information Sheet]	3	31 July 2019
Participant consent form [31.07.19 Generic Participant Consent Form]	3	31 July 2019
Participant consent form [31.07.19 NHS Fife Participant Consent	3	31 July 2019

Form]		
Participant consent form [31.07.19 NHS Lothian Participant Consent Form]	3	31 July 2019
Participant consent form [31.07.19 NHS Forth Valley Participant Consent Form]	3	31 July 2019
Participant consent form [31.07.19 NHS Tayside Participant Consent Form]	3	31 July 2019
Participant information sheet (PIS) [31.07.19 Generic Participant Information Sheet]	3	31 July 2019
Participant information sheet (PIS) [31.07.19 NHS Fife Participant Information Sheet]	3	31 July 2019
Participant information sheet (PIS) [31.07.19 NHS Lothian Participant Information Sheet]	3	31 July 2019
Participant information sheet (PIS) [31.07.19 NHS Forth Valley Participant Information Sheet]	3	31 July 2019
Participant information sheet (PIS) [31.07.19 NHS Tayside Participant Information Sheet]	3	31 July 2019

Approved documents

The final list of approved documentation for the study is therefore as follows:

<i>Document</i>	<i>Version</i>	<i>Date</i>
GP/consultant information sheets or letters [08.07.19 Generic GP and Healthcare Professional Letter]	2	08 July 2019
GP/consultant information sheets or letters [08.07.19 NHS Fife GP and Healthcare Professional Letter]	2	08 July 2019
GP/consultant information sheets or letters [08.07.19 NHS Lothian GP and Healthcare Professional Letter]	2	08 July 2019
GP/consultant information sheets or letters [08.07.19 NHS Forth Valley GP and Healthcare Professional Letter]	2	08 July 2019
GP/consultant information sheets or letters [08.07.19 NHS Tayside GP and Healthcare Professional Letter]	2	08 July 2019
Interview schedules or topic guides for participants [26.10.18 Interview Schedule]		26 October 2018
Interview schedules or topic guides for participants [26.10.18 Interview Schedule]	1	26 October 2018
IRAS Application Form [IRAS_Form_08052019]		08 May 2019
IRAS Application Form XML file [IRAS_Form_08052019]		08 May 2019
IRAS Checklist XML [Checklist_29072019]		29 July 2019
Other [Clinical Trial Liability]	1	31 July 2018
Other [Employers' Liability Insurance]	1	01 August 2018
Other [Indemnity Confirmation]	1	24 July 2018
Other [Professional Indemnity Insurance Confirmation]	1	31 July 2018
Other [26.10.18 Academic Supervisor CV Summary]	1	26 October 2018
Other [08.07.19 Generic Demographic Information Sheet]	2	08 July 2019
Other [08.07.19 NHS Fife Demographic Information Sheet]	2	08 July 2019
Other [08.07.19 NHS Lothian Demographic Information Sheet]	2	08 July 2019
Other [08.07.19 NHS Forth Valley Demographic Information Sheet]	2	08 July 2019

Other [08.07.19 NHS Tayside Demographic Information Sheet]	2	08 July 2019
Other [08.07.19 Generic Participant Debriefing Form]	2	08 July 2019
Other [08.07.19 NHS Fife Participant Debriefing Form]	2	08 July 2019
Other [08.07.19 NHS Lothian Participant Debriefing Form]	2	08 July 2019
Other [08.07.19 NHS Forth Valley Participant Debriefing Form]	2	08 July 2019
Other [08.07.19 NHS Tayside Participant Debriefing Form]	2	08 July 2019
Other [Letter for REC with amendment summary table]	1	22 July 2019
Other [31.07.19 Generic Cystic Fibrosis Nurse Specialist Information Sheet]	3	31 July 2019
Other [31.07.19 NHS Fife Cystic Fibrosis Nurse Specialist Information Sheet]	3	31 July 2019
Other [31.07.19 NHS Lothian Cystic Fibrosis Nurse Specialist Information Sheet]	3	31 July 2019
Other [31.07.19 NHS Forth Valley Cystic Fibrosis Nurse Specialist Information Sheet]	3	31 July 2019
Other [31.07.19 NHS Tayside Cystic Fibrosis Nurse Specialist Information Sheet]	3	31 July 2019
Participant consent form [31.07.19 Generic Participant Consent Form]	3	31 July 2019
Participant consent form [31.07.19 NHS Fife Participant Consent Form]	3	31 July 2019
Participant consent form [31.07.19 NHS Lothian Participant Consent Form]	3	31 July 2019
Participant consent form [31.07.19 NHS Forth Valley Participant Consent Form]	3	31 July 2019
Participant consent form [31.07.19 NHS Tayside Participant Consent Form]	3	31 July 2019
Participant information sheet (PIS) [31.07.19 Generic Participant Information Sheet]	3	31 July 2019
Participant information sheet (PIS) [31.07.19 NHS Fife Participant Information Sheet]	3	31 July 2019
Participant information sheet (PIS) [31.07.19 NHS Lothian Participant Information Sheet]	3	31 July 2019
Participant information sheet (PIS) [31.07.19 NHS Forth Valley Participant Information Sheet]	3	31 July 2019
Participant information sheet (PIS) [31.07.19 NHS Tayside Participant Information Sheet]	3	31 July 2019
Research protocol or project proposal [08.07.19 Study Protocol]	2	08 July 2019
Summary CV for Chief Investigator (CI) [26.10.18 Chief Investigator CV]		26 October 2018

You should ensure that the sponsor has a copy of the final documentation for the study. It is the sponsor's responsibility to ensure that the documentation is made available to R&D offices at all participating sites.

19/SS/0079	Please quote this number on all correspondence
-------------------	---

Yours sincerely

Joyce Clearie
SESREC 2 Manager

E-mail: joyce.clearie@nhslothian.scot.nhs.uk

Copy to: *Ms Blathnaid Greene, NHS Fife*
Dr Amanda Wood, NHS Fife

Lead Nation

Scotland: [nhsg.NRSPCC@nhs.net](mailto:nhsq.NRSPCC@nhs.net)

Appendix F: Ethical Approval from NHS Fife

Medical Director

Hayfield House
Hayfield Road
KIRKCALDY
KY2 5AH



Ms Blathnaid Greene
Trainee Clinical Psychologist
Lynebank Hospital
DUNFERMLINE

Date 11 November 2019
Our Ref 19-037 249098
19/SS/0079
Enquiries to Aileen Yell
E-mail aileen.yell@nhs.net
Telephone 01383 623623 Ext 20940
Website www.nhsfife.org

Dear Ms Greene

Project Title: Parental experiences of receiving a newborn diagnosis of cystic fibrosis

Thank you for your application to carry out the above project. Your project documentation (detailed below) has been reviewed for resource and financial implications for NHS Fife and I am happy to inform you that NHS permission for the above research has been granted on the basis described in the application form, protocol and supporting documentation. The documents reviewed were:

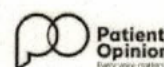
Document	Version	Date
Data Protection Information Sheet	1	24 May 2019
Protocol	2	8 July 2019
GP/Healthcare Professional Letter (NHS Fife)	2	8 July 2019
Interview Schedule	1	26 October 2018
Cystic Fibrosis Nurse Specialist Information Sheet (NHS Fife)	3	31 July 2019
Participant Demographic Information Sheet (NHS Fife)	2	8 July 2019
Participant Information Sheet (NHS Fife)	3	31 July 2019
Participant Consent Form (NHS Fife)	3	31 July 2019
Participant Debriefing Form (NHS Fife)	2	8 July 2019
IRAS R&D Form	5.11	8 May 2019
REC provisional favourable opinion letter		23 June 2019
REC conditional favourable opinion letter		31 July 2019
Study-Wide Governance Report		8 August 2019
REC final favourable opinion letter		28 October 2019
IRAS OID Form		8 November 2019

The terms of the approval state that you are the Principal Investigator authorised to undertake this study within NHS Fife, with assistance from Dr Shona Murphy at Lynebank Hospital, Dunfermline.

I note that the favourable ethical opinion applies to all NHS sites taking part in the study therefore no separate Site Specific Review is required in this case. The sponsors for this study are University of Edinburgh. Please note that it is the responsibility of the Sponsor to ensure that adequate and appropriate insurance is maintained throughout the course of the study.

Details of our participation in studies will be included in annual returns we are expected to complete as part of our agreement with the Chief Scientist Office. Regular reports of the study require to be submitted. Your first report should be submitted to Dr A Wood, R&D Manager, R&D Department, Queen Margaret Hospital, Whitefield Rd, Dunfermline, KY12 0SU in 12 months time and subsequently at yearly intervals until the work is completed. A Lay Summary will also be required upon completion of the project.

⁶ NHS Fife was awarded the Carbon Trust Standard in February 2010 and is the first Scottish NHS Board to achieve this accolade.



In addition, approval is granted subject to the following conditions:-

All research activity must comply with the standards detailed in the UK Policy Framework for Health and Social Care Research http://www.nhsresearchscotland.org.uk/uploads/tiny_mce/uk-policy-framework-health-social-care-research.pdf, health & safety regulations, data protection principles, other appropriate statutory legislation and in accordance with Good Clinical Practice (GCP).

Any amendments which may subsequently be made to the study should also be notified to Aileen Yell, R&D Research Coordinator _____, as well as the appropriate regulatory authorities. Notification should also be given of any new research team members post approval and/or any changes to the status of the project.

This organisation is required to monitor research to ensure compliance with the Research Governance Framework and other legal and regulatory requirements. This is achieved by random audit of research. You will be required to assist with and provide information in regard to monitoring and study outcomes (including providing recruitment figures to the R&D office as and when required).

As custodian of the information collated during this research project you are responsible for ensuring the security of all personal information collected in line with NHS Scotland IT Security Policies, until the destruction of this data. Permission is only granted for the activities for which a favourable opinion has been given by the REC (and which have been authorised by the MHRA where appropriate).

The research sponsor or the Chief Investigator or local Principal Investigator at a research site may take appropriate urgent safety measures in order to protect research participants against any immediate hazard to their health or safety. The R&D office (aileen.yell@nhs.net) should be notified that such measures have been taken. The notification should also include the reasons why the measures were taken and the plan for further action. The R&D office should be notified within the same time frame of notifying the REC and any other regulatory bodies.

I would like to wish you every success with your study and look forward to receiving a summary of the findings for dissemination once the project is complete.

Yours sincerely

A small, handwritten signature in dark ink, appearing to be 'DR CHRIS MCKENNA'.

DR CHRIS MCKENNA
Medical Director
NHS Fife

*Cc : Aileen Yell, R&D Research Coordinator, NHS Fife, Queen Margaret Hospital, Dunfermline
Dr Shona Murphy, NHS Fife*

Appendix G: Ethical Approval from NHS Lothian

University Hospitals Division

Queen's Medical Research Institute
47 Little France Crescent, Edinburgh, EH16 4TJ

FM/AM/approval

20th December 2019

Ms Blathnaid Greene
NHS Fife
Lynebank Hospital
Halbeath Road
Dunfermline
KY11 4UW



Research & Development
Room E1.16
Tel: 0131 242 3330

Email:
accord@nhslothian.scot.nhs.uk

Director: Professor Tim Walsh

Dear Ms Greene

Lothian R&D Project No: 2019/0168

REC No: 19/SS/0079

Title of Research: Parental experiences of receiving a newborn diagnosis of cystic fibrosis.

Sponsor Reference: CAHSS1808/01

Participant Information Sheet:

Version 3, dated 31st July 2019
(Data Protection) Version 1, dated 24th May 2019
(Nurse Specialist) Version 3, dated 31st July 2019
(Demographic) Version 2, dated 8th July 2019
(Debriefing form) Version 2, dated 8th July 2019

Consent Form:

Version 3, dated 31st July 2019
(Data Protection) Version 1, dated 24th May 2019

Protocol: Version 2, dated 8th July 2019

I am pleased to inform you this letter provides Site Specific approval for NHS Lothian for the above study and you may proceed with your research, subject to the conditions below.

Please note that the NHS Lothian R&D Office must be informed of any changes to the study such as amendments to the protocol, funding, recruitment, personnel or resource input required of NHS Lothian.

Substantial amendments to the protocol will require approval from the ethics committee which approved your study and the MHRA where applicable.

Please keep this office informed of the following study information, **which is a condition of NHS Lothian R&D Management Approval:**

1. Date you are ready to begin recruitment, date of the recruitment of the first participant and the monthly recruitment figures thereafter.
2. Date the final participant is recruited and the final recruitment figures.
3. Date your study / trial is completed within NHS Lothian.

I wish you every success with your study.

Yours sincerely

Ms Fiona McArdle
Deputy R&D Director

Appendix H: Participant Information Sheet (logos adapted to health board)



PARTICIPANT INFORMATION SHEET

You are being invited to take part in a project. We hope the information provided in this information sheet will help you decide if you are interested in participating. Please read it carefully. Thank you.

What is the purpose of the study?

Receiving a health diagnosis for your child can evoke a range of responses from parents. This study is interested in finding out about parents' experiences of their child receiving a diagnosis of cystic fibrosis (CF) following newborn screening. We are keen to learn about this process from parents' perspectives and what they think would be useful for future families receiving the diagnosis. We hope that by gathering parents' views we can better understand their experiences, adding knowledge to this under-researched area and consider ways in which services can be delivered.

This study is also part of the principal researcher's Doctorate in Clinical Psychology degree.

Why have I been invited?

You have been invited to participate via your CF service. We are inviting parents who have a child who has been diagnosed with CF at least 6 months ago following newborn screening in Scotland.

Do I have to take part?

No. There is no obligation to take part. It is entirely your choice whether you would like to participate or not. If you do decide to take part you will be given this information sheet to keep and be asked to sign a consent form. If you decide to take part you are still free to withdraw at any time. Your decision to take part or withdraw from the study will not impact on the care your family receives. If you have any questions, you are welcome to discuss these with your CF nurse specialist or the principal researcher.

What will happen if I take part?

If you decide to take part in the study, the researcher will arrange a time to meet that is convenient for you. They will go through the consent form with you and answer any questions you may have. If you are still happy to take part, you will be asked to sign the consent form. We will also ask your permission to inform your CF nurse and/or your GP that you are taking part, in case there is anything you may want to discuss with them. We will not share your responses with them, simply inform them that you are taking part.

You will meet with the researcher for approximately 30-90 minutes, this depends on how much you are comfortable sharing. You do not have to answer any questions you do not want to and you can stop the interview at any point. At the end of this information sheet there are examples of the types of questions you might be asked.

The researcher will record the conversation using a Dictaphone and will type up the conversation afterwards. Once it has been typed, the recording will be destroyed. The typed text will be used to analyse information that parents share. All of the information will be kept safely according to National Health Service (NHS) policies. At the end of the meeting, you will have a chance to discuss your experience of the interview with the researcher.

You will also be asked if you would like a summary of the findings from the research once the study is complete. If you would like to receive this information, we can send it to you via email or in the post. The findings from the research will be part of the researcher's doctoral thesis in Clinical Psychology and it is hoped that they will also be published. No identifiable information will be included in these.

What are the possible disadvantages or risks of taking part?

The interview may involve discussing difficult experiences though you do not need to discuss anything you do not wish to. The topics discussed during the meeting could potentially be upsetting. If you become upset, you will have the opportunity to discuss this with the researcher and/or your CF nurse specialist and GP. We hope that by including examples of the sorts of questions you will be asked you will have an idea of what to expect before taking part.

What are the possible benefits or advantages of taking part?

One possible benefit might include having the opportunity to discuss your experience with someone outside of your usual family/friends/healthcare providers in a safe, non-judgemental and empathic environment. Another potential benefit may be the opportunity to contribute to improving knowledge and understanding of parents in this situation. It is hoped that the information gathered during this study may have an impact on service delivery for future families.

Will my taking part in the study be kept confidential?

The information you provide will be kept confidential. All recordings will be kept on encrypted recorders and any forms you sign or complete will be kept in a locked filing cabinet on NHS premises, in line with NHS policies. Your data will be anonymised and a code will replace your name. Only members of the research team will have access to identifiable information.

The principal researcher is a member of the Protecting Vulnerable Groups (PVG) scheme. The only time confidentiality would be broken is if the researcher was concerned for your safety or the safety of someone else (e.g. a child). In these rare cases, local area and NHS child protection disclosure processes will be followed (more information on this can be found, for example, here: <https://www.fifedirect.org.uk/minisites/index.cfm?fuseaction=page.display&pageid=5C8A3FA2-F091-A0B3-6CA6A35C4DCE744F&siteID=AA73CD9C-E7FE-C7EA-06436BFC786E1C8E>). If we did need to break confidentiality, we would do our best to inform you of this beforehand and keep you updated.

The CF nurse specialists and your GP (if you have provided permission) will be aware of who has been invited to participate in research. They will not be provided with details of the content of the interview but will be available if you need further support. When the study is written up no identifiable information will be included. Quotes from the conversations may be included word for

word in the write-up but these will not be linked to any information about specific parents and pseudonyms will be used.

What if I have questions?

If you have any questions you can contact the principal researcher, Bláthnaid Greene. She can be contacted via telephone on 01383 565 400, by email: blathnaidgreene@nhs.net or in writing: Bláthnaid Greene, Clinical Psychology Department, Lynebank Hospital, Dunfermline KY11 4UW.

If you would like to discuss this study with someone independent of the study team please contact: Tara Graham, Research & Service Development Psychologist, on 01334 696 336 or taragraham@nhs.net.

What if I want to take part?

If you would like to take part, please let your CF nurse specialist know and they can pass on your details or contact **Bláthnaid Greene** directly on **01383 565 400** or by email:

We look forward to hearing from you.

Thank you very much 😊

Bláthnaid Greene, Principal Researcher, The University of Edinburgh and NHS Fife
Dr Shona Murphy, Clinical Psychologist, NHS Fife
Dr Corinne Reid, Reader in Clinical Psychology, The University of Edinburgh
Dr Mark Hoelterhoff, Lecturer in Clinical Psychology, The University of Edinburgh

Examples of the types of questions you might be asked:

- What do you recall about receiving the diagnosis?
- How did you receive the diagnosis? What was the experience like for you?
- Were there any parts of how you received the diagnosis that you felt were helpful/unhelpful?
- What impact did receiving the diagnosis have on you/your family?
- What do you feel would be helpful for families receiving a CF diagnosis following newborn screening?

Feedback, Concerns or Complaints:

If you wish to provide feedback or make a complaint you can contact NHS Fife's Patient Relation Department on 01592 648 153; Patientrelations.fife@nhs.net; or in writing to:

Patient Relations Department
Fife NHS Board
Room 104
Hayfield House
Hayfield Road
Kirkcaldy
KY2 5AH

You can find out more about how we use your information and our legal basis for doing so in our Privacy Notice at www.accord.scot.

For further information on the use of personal data by NHS sites, please link to the Health Research Authority (HRA) website; <https://www.hra.nhs.uk/information-about-patients/>.

If you wish to raise a complaint on how we have handled your personal data, you can contact our Data Protection Officer who will investigate the matter. If you are not satisfied with our response or believe we are processing your personal data in a way that is not lawful you can complain to the Information Commissioner's Office (ICO) at <https://ico.org.uk/>.

Data Protection Officer contact information:

University of Edinburgh
Data Protection Officer
Governance and Strategic Planning
University of Edinburgh
Old College
Edinburgh
EH8 9YL
Tel: 0131 651 4114
dpo@ed.ac.uk

Appendix I: Cystic Fibrosis Nurse Specialist Information Sheet (logos adapted to health board)



CYSTIC FIBROSIS NURSE SPECIALIST INFORMATION SHEET

Thank you very much for your willingness to support us with this research project. We hope this information sheet will be useful, please let us know if you have any further questions.

What is this study about?

We are hoping to interview between 8-12 parents whose child has received a diagnosis of cystic fibrosis (CF) following newborn screening. We are interested in finding out about parents' experiences of receiving the diagnosis, the impact the diagnosis has had on them and what they feel may be useful for future families. This appears to be an under-researched area and we are keen to gain a better understanding of parents' experiences.

Inclusion criteria:

- Parent of a child with CF
- Adult, aged 18 years or older
- Participant able to provide informed consent
- Child's CF diagnosis received from newborn screening
- Child's diagnosis received at least 6 months prior to interview
- Parent participants have remained primary caregivers for their child

Exclusion criteria:

- Child's diagnosis received less than 6 months ago
- Child's diagnosis received outside of Scotland
- More than one child with cystic fibrosis in family

What do I need to do?

We would be very grateful if you can provide appropriate parents with the "Participant Information Sheet" and, with parents' permission, pass on the details of anyone who might be interested to Bláthnaid Greene (blathnaidgreene@nhs.net). Parents are also welcome to think about taking part and contact Bláthnaid directly.

We will of course be happy to provide any further information. If parents decide they are willing to participate, Bláthnaid will arrange interview times with parents directly.

Aside from sharing information between parents and the research team, we do not expect that any further input will be needed. We recognise the hugely valuable role you play in the care of these

Parental experiences of newborn diagnosis of CF

v2 08.07.2019
1 of 2

families and the importance of your existing relationship with parents. As such, the only other time we may contact you regarding parents is if families may require some additional support following the interview.

What is involved for the parent?

Bláthnaid will meet with parents for one semi-structured interview. Before the interview begins, she will answer any questions and obtain parents' consent to participate. It will be made clear that participants do not need to answer any questions they do not want to and they can stop the interview at any point. Confidentiality will be explained and agreed to before the interview commences. Depending on how much information parents are willing to share and/or how comfortable they are, the interviews will last between 30-90 minutes.

Though these interviews are semi-structured and the researchers have some guiding questions in mind, we are interested in parents' experiences and will therefore be guided by what parents decide to discuss. The conversation will be recorded using a Dictaphone and the interviews will later be transcribed. All information will be stored in line with NHS Fife policies.

Once the research is completed, full reports and a summary of the findings will be made available to all participants and CF services who wish to receive this information.

Please do not hesitate to contact us if you have any further questions.

Thank you again for your help, we greatly appreciate it.

Bláthnaid Greene, Principal Researcher, The University of Edinburgh and NHS Fife

Telephone: 01383 565 400

Email: blathnaid.greene@nhs.uk

Dr Shona Murphy, Clinical Psychologist, NHS Fife

Telephone: 01334 696 336

Email: shona.murphy@nhs.uk

Dr Corinne Reid, Reader in Clinical Psychology, The University of Edinburgh

Telephone: 0131 650 4270

Email: corinne.reid@nhs.uk

Dr Mark Hoelterhoff, Lecturer in Clinical Psychology, The University of Edinburgh

Email: mark.hoelterhoff@nhs.uk

Appendix J: Participant Consent Form (logos adapted to health board)



PARTICIPANT CONSENT FORM



Title of Project: Parental experiences of receiving a CF diagnosis following newborn screening

Name of Researchers: Bláthnaid Greene, Dr Shona Murphy, Dr Corinne Reid, Dr Mark Hoelterhoff

<i>Participant ID:</i>		Please <u>initial</u> each box
1.	I confirm that I have read and understood the participant information sheet (v3 31.07.19). I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily.	
2.	I understand that my participation is voluntary and that I am free to withdraw at any time, without giving any reason, without my medical care or legal rights being affected.	
3.	I understand that my participation in this project will involve taking part in an interview with the researcher and completing a short questionnaire.	
4.	I agree to my interview being recorded on a digital audio recorder.	
5.	I understand that data collected during this study may be looked at by other members of staff at the University of Edinburgh and NHS Fife who are linked to the research (e.g. researcher supervisors). I give them permission to have access to my data.	
6.	I understand and agree that some direct quotations from my interview may be used within the researcher's doctoral thesis and future publications that arise from this research. I understand that all quotations will be anonymised.	
7.	The limits of confidentiality have been explained to me and I understand that if any issues relating to person safety or other risks are raised during the interview, the researcher will follow relevant NHS Health Board and University of Edinburgh protocols to person safety.	
8.	I give permission for a member of the CF team to be notified that I am taking part in the research.	
9.	I give permission for my GP to be notified that I am taking part in the research.	
10.	I understand that relevant sections of data collected during the study may be looked at by individuals from the regulatory authorities and from the Sponsor (the University of Edinburgh) or from the/other NHS Board(s) where it is relevant to my taking part in this research. I give permission for those individuals to have access to my records.	
11.	I agree to be contacted again to discuss the interpretations and conclusions drawn from my interview.	
12.	I would like to be contacted again with information on the findings of this research (e.g. summary of the findings).	

13.	I agree to take part in the above study.	
-----	--	--

Name of Participant

Date

Signature

Name of Person
taking consent

Date

Signature

Original (x1) to be retained in site file. Copy (x1) to be retained by the participant.

Appendix K: Participant Debriefing Form (logos adapted to health board)



DEBRIEFING FORM



Title of Project: Parental experiences of receiving a cystic fibrosis (CF) diagnosis following newborn screening

Name of Researchers: Bláthnaid Greene, Dr Shona Murphy, Dr Corinne Reid, Dr Mark Hoelterhoff

Thank you very much for participating in this research, we greatly value your time and input. The information gathered from the interviews conducted during this study will help us better understand the experiences of parents who have received a CF diagnosis following newborn screening. As well as increasing understanding of these experiences, it is hoped that findings will contribute to service delivery for future families. If you feel you may need any additional support, please find a list of support services on the back of this form. You are also welcome to discuss any issues with your CF team or GP.

It is important to the research team, that the conclusions we draw from the interviews are fair interpretations of the interviews. As such, we would like to speak to parents who have taken part to hear their input on these interpretations and whether they are accurate representations of what parents have shared with us. It is estimated that this would take approximately 30 minutes and could be done in person or by telephone. Should you be willing to do this, I would contact you in a few months to set up a convenient time to review the interpretations. This is completely voluntary and there is no obligation to take part in this additional stage. You may change your mind at any time and, as with the main study, your decisions will not impact the care your family receives in any way.

The findings from this study will be submitted as part of my clinical psychology doctoral thesis. It is hoped the findings will also be shared in scientific journals, conferences and feedback sessions with the CF services that have taken part. It is common for some quotations from interviews to be included when results are shared. If you have given permission, quotations from your interview may be used. Importantly, these will all be anonymised and it will not be possible to identify individual participants.

If you would like to receive a summary of the findings of this research, please confirm your contact details with Bláthnaid.

Thank you again.

Bláthnaid Greene, Trainee Clinical Psychologist

Psychology Department, Lynebank Hospital, Dunfermline, Fife Telephone: 01383 565 400

SUPPORT SERVICES

It is not thought that participating in this study will result in any serious or long-term psychological harm. We recognise, however, that some of the topics discussed or thought about during this study may be upsetting for some participants. If you feel you may need some additional support please discuss any issues with a member of your CF team or your GP.

Please find below details of support services that may also be able to provide additional support.

<p>SAMARITANS</p> <p>Emotional support to anyone in emotional distress, struggling to cope, or at risk of suicide.</p> <p>Telephone: 116 123 Website: https://www.samaritans.org/</p>	<p>NHS 24</p> <p>Comprehensive health information and self-care advice to the people of Scotland.</p> <p>Telephone: 111 Website: https://www.nhs24.scot/</p>
<p>BREATHING SPACE</p> <p>Free, confidential service for people in Scotland experiencing low mood, depression or anxiety.</p> <p>Telephone: 0800 838 587 Website: http://breathingspace.scot/</p>	<p>CHILDLINE</p> <p>Counselling service for children and young people up to their 19th birthday.</p> <p>Telephone: 0800 1111 Website: https://www.childline.org.uk/</p>

Complaints Procedure:

We hope taking part in this research has met your expectations. If you would like to make a complaint about the study, please contact the University of Edinburgh's Research Governance team (researchgovernance@ed.ac.uk). The team will be able to guide you through the complaints' procedure process.

If you wish to provide feedback or make a complaint you can contact NHS Fife's Patient Relation Department on 01592 648 153; Patientrelations.fife@nhs.net; or in writing to:

Patient Relations Department
Fife NHS Board
Room 104
Hayfield House
Hayfield Road
Kirkcaldy
KY2 5AH

Appendix L: Semi-Structured Interview Schedule

INTERVIEW SCHEDULE

Principles of interviewing: Interested in participant and their experience. No right or wrong answers. Some questions may be self-evident, trying to get to grips with how participant understands things. Can take time and think. Participant as expert in their own experience. Active listening. Rapport-building.

1. Can you tell me what life was like for you (and your family) before you received the diagnosis?
2. Can you tell me what you recall about receiving the diagnosis?
3. Can you tell me how you received the diagnosis? What was the experience like for you?
4. Are there any parts of how you received the diagnosis that you felt were helpful/unhelpful?
5. Can you tell me what impact receiving the diagnosis (in the way you did) has had on you/your family?
6. What do you feel would be helpful for families receiving a CF diagnosis following newborn screening?

Appendix M: Study Protocol



STUDY PROTOCOL

Study: Parental experiences of receiving a newborn diagnosis of cystic fibrosis.

Protocol Author: Bláthnaid Greene, Principal Researcher and Trainee Clinical Psychologist

Supervisors: Academic – Dr Corinne Reid and Dr Mark Hoelterhoff, University of Edinburgh
Clinical – Dr Shona Murphy, NHS Fife

REC reference number: 19/SS/0079

BACKGROUND AND RATIONALE

Overview of Cystic Fibrosis and the Scottish Context

Cystic fibrosis (CF) is a genetically inherited condition affecting over 10,400 people in the United Kingdom (Cystic Fibrosis Trust, 2018) and around 20 newborns every year in Scotland (Scottish Government, 2015). This life-limiting condition affects mainly the respiratory and digestive systems due to the build-up of thick sticky mucus impacting functioning and increases individuals' susceptibility to infections (National Health Service UK, 2018a; The Butterfly Trust, 2018). Significant improvements in survival with CF have been observed since the introduction of specialised CF care (Smyth et al., 2014). Part of this specialised care has been the implementation of newborn heel prick blood spot screening which includes screening for CF as well as other hereditary conditions (National Health Service UK, 2018b). The national newborn screening programme for CF began in Scotland in 2003 (Scottish Government, 2015).

Impact of Cystic Fibrosis on Families

Chronic health conditions, physical and psychological, have been recognised as increasing the risk of emotional distress throughout the family (Holmes & Deb, 2003). Having a child with CF may have a variety of biological, social and psychological impacts on families. Treatment regimens can be complex and very time-consuming, including taking a range of medications and receiving physiotherapy or physical interventions (Glasscoe & Quittner, 2008). Finding out a genetic condition exists within the family can bring additional responsibilities impacting future decision-making and information-sharing with the wider family system (Parsons & Bradley, 2003).

Reviewing qualitative studies of children and young peoples' experiences of CF, Jamieson and colleagues (2014) note that the effects of living with CF impact on identity, daily functioning (in part due to physical health constraints and infection control) and life goals. Reporting on the sense of "vulnerability, loss of independence and opportunities, isolation and disempowerment" (p.1683) described by participants in the forty-three articles reviewed, the authors emphasise the importance

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of multi-disciplinary, person-centred care. Systematically reviewing literature from the preceding two decades, Berge and Patterson (2004) report that as well as impacts on the individual with CF, psychosocial impacts have been found in siblings and parent caregivers.

Given the additional stressors and complexities faced by families with a child with CF it is perhaps unsurprising that this population may be at higher risk of psychological difficulties. Examining responses to screening tools for anxiety and depression from young people and adults with CF and parent caregivers in a multi-country epidemiological study, Quittner and colleagues (2014) report elevation rates two to three times that of community samples. Alongside the overall elevation in symptoms of anxiety and depression across the participants, Quittner et al. examined dyads with parents and adolescent reports. These analyses found adolescents were significantly more likely to be above clinical cut-offs if their parents' responses were elevated.

The importance of caregivers to children with chronic health conditions, their quality of life and wellbeing has been highlighted in the literature (for example, Chow, Morrow, Cooper Robbins, & Leask, 2013; Cousino & Hazen, 2013). Parents of children with CF are required to take on additional caregiving roles such as treatment administration and management which can come with a range of financial, practical and emotional challenges (Fitzgerald, George, Somerville, Linnane, & Fitzpatrick, 2018). These new roles may result in a change in personal identity for parents and an increase in "burden of responsibility" (Hodgkinson & Lester, 2002) which often occur alongside uncertainty and changes in children's health status (Fitzgerald et al., 2018). Being a mother, increasing child age and ever having a positive *Pseudomonas aeruginosa* result (indicative of disease severity) have been found to be associated with greater reports of parental burden (Fitzgerald et al., 2018). Higher parental stress is associated with lower psychological adjustments in both parents and children (Cousino & Hazen, 2013). Poorer treatment adherence for children has been found with higher levels of parental stress (Eddy et al., 1998) and parental depression (Quittner, Barker, Geller, Butt, & Gondor, 2007; Smith, Modi, Quittner, & Wood, 2010). This emphasises the need to consider parent caregivers at all stages to promote wellbeing across the family.

Neonatal Screening for Cystic Fibrosis

Benefits linked to neonatal screening for CF include, reduced disease severity, burden of care and costs (Castellani et al., 2009; Southern, Mérelle, Dankert-Roelse, & Nagelkerke, 2009), increasing opportunities for individuals to live with greater quality of life and life expectancy (Smyth et al., 2014). Within the European best practice guidelines for CF neonatal screening, risks identified include carrier status disclosure and diagnostic uncertainty and the need for clear communication with families is highlighted (Castellani et al., 2009). International standards of care and best practise guidelines provide biological and medical information for diagnosis as well as information describing the "minimum acceptable standards for reporting a CF diagnosis following newborn screening to families" (Smyth et al., 2014, p.27).

Whilst this may emphasise the importance of information-sharing, the guidelines are limited to three criteria (results should be discussed with a CF specialist, families should receive written information to refer to later and families should have clear understanding for short and long term management plans). Studies with families who have gone through the screening process report increased levels of anxiety and distress and though this may be more transient depending on the outcome of screening, the experience appears to be shared (Hayeems et al., 2016; Parsons & Bradley, 2003; Ulph, Cullinan, Qureshi, & Kai, 2015).

Experiences of Cystic Fibrosis Diagnosis and Newborn Screening

Receiving the news that your child has CF is the beginning of a long-term process and appears to be a salient memory for parents (Chudleigh et al., 2016; Havermans, Tack, Vertommen, Proesmans, & de Boeck, 2015). Parents' favourable attitudes to newborn screening have been taken as an indicator of the acceptability of early diagnosis (Parsons & Bradley, 2003). Whether families have received diagnosis through newborn screening or later on in childhood, newborn screening appears to be viewed as advantageous by parents (Helton, Harmon, Robinson, & Accurso, 1991 in Parsons & Bradley, 2003).

Recognising the minimal amount of evidence investigating parents' experiences of receiving a positive newborn screening result, Chudleigh and colleagues (2016) explored such experiences for CF and sickle cell disease in England. Using a grounded-theory approach, six themes were identified and contributed to a proposed theoretical framework exploring factors that impact on parents when receiving a positive result. Parents who received a result for CF generally had limited knowledge of the condition which added increased anxiety to other emotional reactions (e.g. guilt, denial, shock). Social factors, including sharing the results with others and difficulties in parental relationships, were also reported as important experiences by parents.

The professional delivering the results and the way in which information was shared had a substantial impact on parents. Overall, a face-to-face meeting with a CF specialist and both parents seemed to be the favoured approach. Though parents were advised not to use the internet to find additional information in the short-term, parents reported using this as an information-gathering strategy. The concepts of information and education were specifically explored in parents receiving care from an Australian hospital which offers follow-up multidisciplinary education (Jessup, Douglas, Priddis, Branch-Smith, & Shields, 2016). Parents in this study recalled initially feeling overwhelmed and unable to assimilate all information. Similarly to participants in Chudleigh et al.'s (2016) study, parents generally had little prior CF knowledge and experienced shock and disbelief. Whilst some consensus appears to exist for issues such as the setting of diagnosis delivery and professional delivering information, the amount, timing and staggering of information present a more mixed picture (Jessup et al., 2016). Whilst some wanted as much information as possible straight away, others felt less information and more comforting was needed. This variance is reflective of the complex processes involved in receiving such a diagnosis and perhaps emphasises the need for professionals delivering this information to be flexible and attuned to the needs of individual family members.

Research Rationale and Implications

Receiving the diagnosis of CF is a significant and potentially distressing life event for families. When diagnosis is confirmed from newborn screening it is occurring during the perinatal period which is already recognised as a period of 'unparalleled change' for families (Howard, Piot, & Stein, 2014). Parents' experiences of receiving the diagnosis may have an impact on how they cope and adjust which can have family-wide implications. Initial quantitative data collected on parents' experiences of receiving a CF diagnosis following newborn screening in Scotland (Smith & Murphy, unpublished) show parallels with some of the research discussed above with respondents choosing to provide additional written information following some questions. This may indicate a willingness and desire for parents to share their story of this process. The current study aims to explore these experiences for parents who have received the diagnosis in Scotland. It will also add to the small body of research in this area by using a different approach (IPA) and by considering the context in which families have found themselves prior to receiving the diagnosis, an area which does not yet appear to have been explored. It is hoped that findings from this research will help inform service delivery as well as contributing to the literature in this area.

RESEARCH QUESTIONS

Principal

What are parents' experiences of receiving a newborn diagnosis of cystic fibrosis?

Secondary

What were families' contexts/lives like before receiving the diagnosis?

What do parents recall about receiving the diagnosis?

What impact has receiving the diagnosis had on families?

How would families like to receive this diagnosis information?

METHODOLOGY

Design

As this study aims to explore the *experience* of parents in relation to a specific event, it will employ a qualitative phenomenological research design, an approach recognised for capturing the lived experiences of groups of people (Chenail, 2011; Finlay, 2011). Since Interpretative Phenomenological Analysis (IPA) is "concerned with the detailed examination of personal lived experience, the meaning of experience to participants and how participants make sense of that experience" (Smith, 2011, p.9) it is viewed as a suitable approach for exploring the research questions of this project. Guidelines specific to IPA proposed by Smith (2011) will be followed.

Participants

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Participants will be parents of children who have been diagnosed with CF through newborn screening in Scotland. Recognising that fathers are often underrepresented in research (Chudleigh et al., 2016; Phares, Lopez, Fields, Kamboukos, & Duhig, 2005), both mothers and fathers will be invited to participate. Families will already be linked in to a CF centre in Scotland from which their child receives their CF-related care. This study aims to interview between 8 and 12 participants.

Inclusion criteria:

- Parent of a child with CF
- Adult, aged 18 years or older
- Participant able to provide informed consent
- Child's CF diagnosis received from newborn screening
- Child's diagnosis received at least 6 months prior to interview
- Parent participants have remained primary caregivers for their child

Exclusion criteria:

- Child's diagnosis received less than 6 months ago
- Child's diagnosis received outside of Scotland
- More than one child with cystic fibrosis in family

Recruitment

Participants will be recruited from National Health Service (NHS) paediatric CF centres and outreach centres in Scotland. Families receive multidisciplinary (MDT) input from professionals in these centres and these pre-existing relationships are hoped to help with this study in several ways. Firstly, by having professionals who can explain any further questions participants may have regarding the research or put them in touch with the principal researcher. Secondly, by providing additional support and managing any risk that may become apparent during the research. Recruitment will focus on South East of Scotland CF services.

Measures

Participants will be interviewed in person using a semi-structured interview guide. This interview schedule will be created based on the research questions outlined above exploring parents' experiences of receiving CF diagnosis following newborn screening. An 'expert-by-experience' (parent of a child with CF) will be asked to review and comment on the questions and provide input regarding interview practicalities, for example, whether they think these should take place one-to-one or in the form of a focus group. A professional from one of the CF centres will also be invited to comment at this point. It is hoped that taking stakeholder perspectives in to account at this stage will enrich the overall research.

In line with IPA guidelines, questions will be open-ended and flexible to allow exploration of topics raised by parents. Interviews are estimated to take between 30-90mins. All interviews will be digitally recorded and held in accordance with local NHS Fife information governance.

Procedure

Input from stakeholders

In the first instance, key stakeholders will be invited to provide input. A parent expert-by-experience will be contacted to review questions and asked to provide any additional comments on issues. MDT CF service professionals will also be asked for their input.

Information for CF services and participant selection

The clinical supervisor for this project is part of the NHS Fife paediatric CF service and, along with the researcher, will provide information to the broader CF service team members regarding the research project and recruitment. Participants meeting inclusion criteria will be identified through the service and invited to take part in the research.

Parents who are interested will be provided with participant information outlining the rationale for the research and what may be involved in taking part. Those who would like to take part or find out more information can contact the researcher directly or give the CF clinician permission to pass their details on to the researcher. The principal researcher will communicate with the participants to set up a meeting for the interview and will be able to ask any questions they may have. It will be made clear that parents are under no obligation to take part, that they can withdraw at any time and that this will not impact on the care their family receives.

Interview

Written consent will be obtained before the interview and demographic information will be collected. One-to-one interviews will either take place in participants' homes or NHS settings. In line with NHS guidelines, confidentiality and its limits will be made explicit and lone worker policies will be followed.

Following qualitative recommendations (Smith, Flowers, & Larkin, 2009), the researcher will aim to establish rapport with participants, provide a non-judgemental space and be an active listener. It is hoped that this will encourage parents to speak freely and openly about their experiences. The questions will be used as a guide but the researcher will be responsive to information shared by participants and be guided by what is shared within each interview.

Post interview

Participants will have the opportunity to debrief following the interview. They will also be provided with details of support services in case they would like to contact them in the future. Participants will be reminded of the professionals involved in their care (e.g. CF MDT, GP) with whom they can speak if they wish to discuss anything that has come up in the interview further. Paediatric CF specialist nurses will be aware of the research taking place and will be a continued source of support for families.

It is hoped that at least some participants will be willing to review interpretations of the data once it has been analysed. Permission will be sought to contact participants if they are happy to be contacted for this purpose and/or if they would like to be informed of the findings of this research.

Ethical considerations

In terms of ethical considerations during research, given the nature of the topic being explored, it is possible that this could be emotive for participants. As a trainee clinical psychologist, the researcher has experience of working with people who may show signs of distress and will be able to monitor appropriately for this. With this population, one substantial protective factor is that families are linked in to and monitored by a CF clinical team. The existing relationships they have with these teams will act as an additional source of support if needed.

The researcher will receive academic and clinical supervision from the listed supervisors throughout this process which will enable reflection on and discussion of any arising issues.

DATA COLLECTION

Demographic data

Demographic data specific to the parent participant (i.e. age, nationality, relationship and employment status) will be collected prior to the interview. Information relating to their child and wider family will also be collected, including, number of children and ages, date when CF diagnosis was delivered, health board where diagnosis was received, other diagnoses within the family, family structure (e.g. who lives at home).

Measures

Guidance on IPA data collection provided by Smith, Flowers and Larkin (2009) will be followed. In line with this, a schedule consisting of six to ten questions will be created for semi-structured interviews with participants. The schedule will be reviewed by stakeholders and will aim to provide parents with some direction for discussing their experiences of receiving a newborn diagnosis of CF whilst remaining open-ended and broad enough for parents to raise and discuss other issues.

Interviews

Interviews will be guided by the schedule, recognising that not all of the questions need to be asked or presented in the same order for each participant (Smith et al., 2009). A strength of phenomenological approaches is their ability to bridge research and clinical practise (Finlay, 2011). The practitioner researcher can make use of their clinical interviewing, active listening and reflective skills and this will be the aim of the primary researcher during these interviews. The researcher will also be mindful of their ethical responsibilities and will monitor the effect of the interview on the participant attentively, responding to this as necessary (e.g. paying attention to non-verbal behaviour, rephrasing etc.) (Smith et al., 2009). As discussed above, the schedule will be used flexibly to make way for any unexpected topics to be explored. Interviews will be recorded using NHS digital recorders, following NHS and University of Edinburgh guidance on recording sessions with clients.

Pilot Interview

The first interview will be used to review the schedule and interview strategies (Smith et al., 2009) and obtain feedback from the participant. This will be discussed with supervisors to consider how best to proceed with following interviews. The decision to include or exclude the first interview in the final data analysis will depend on whether substantial changes were made to the interview process; again, reflection with supervisors will guide this decision.

SAMPLE SIZE

There is a consensus, for example within the field of health psychology research, that IPA uses smaller sample sizes (Brocki & Wearden, 2006) with a focus on depth of experience (Smith et al., 2009). IPA has been utilised as research approach investigating parental experiences of their child having different health conditions such as severe food allergies (Rouf, White, & Evans, 2012) and cancer (Schweitzer, Griffiths, & Yates, 2012). These studies used sample sizes of 8 and 11 parents respectively.

Examining research that has employed qualitative approaches to look at parental experiences following a newborn screening CF diagnosis, two studies have used a sample size of 10. One of which employed a van Manen's phenomenological approach (Jessup et al., 2016) and the other grounded theory (Chudleigh et al., 2016). Though the Chudleigh and colleagues' study (2016) had a total of 22 participants, 12 were parents of children with sickle cell disease with the remaining 10 being parents of children with CF.

Taking in to consideration some of the theoretical discussion on sample sizes for IPA as well as previous studies in areas similar to those being investigated in this study, it is estimated that a sample size between 8-12 participants will be sufficient to address the study's aims.

ANALYSIS

Within IPA, the focus of analysis is placed upon "participants' attempts to make sense of their experiences" (Smith et al., 2009, p.79). Interviews will be transcribed and anonymised by the primary researcher. The process of analysis described by Smith and colleagues (2009) will be followed. To begin, each interview will be read and re-read whilst listening to the recording for the

researcher to be familiar and immersed in the material. Initial note-taking and underlining/highlighting will be done in which the researcher will endeavour to “maintain an open mind and note anything of interest within the transcript” (p.83). As well as increasing familiarity with the content, this will produce detailed notes and comments. Comments at this stage may fall in to a variety of categories, including descriptive, linguistic and conceptual.

Following this process of engagement with the material, the researcher will identify emergent themes, working from the initial transcripts and notes taken. Emergent themes will then be grouped together based on similarities or relatedness to create super-ordinate themes. This will be done for each of the interviews. Once this process has been completed from the data for each participant, the researcher will look for patterns across cases. During this process it is hoped that shared qualities will be evident across cases and there may also be more individual or unique themes. A master table of themes will then be created for the entire study to be explored for deeper levels of interpretation.

The primary researcher will review and discuss the data analysis and interpretation with both supervisors throughout this process. A blind analysis of one of the interviews will be done with at least one of the supervisors to compare and review themes identified. This is done with the aim of increasing validity and credibility by taking additional perspectives in to account. Another important part of increasing validity is offering participants the opportunity to provide feedback on the interpretation of the data. As detailed above, participants will be invited to review the interpretations.

A reflective journal will be kept throughout the research process to keep note of the primary researcher’s experiences, thoughts and assumptions. The procedure described above will also be recorded to evidence the analysis process.

DATA MANAGEMENT AND CONFIDENTIALITY

Participant contact details (name and contact number) provided by a clinician of the CF paediatric service will be stored on a password protected NHS Fife computer located at the Chief Investigator’s main base, Lynebank Hospital, Dunfermline, Fife. Hard copies of participant consent forms, and demographic forms will be stored in a secured cabinet at the Chief Investigator’s main base, this will not be accessible to members of the public. The key linking participants names to unique identifier codes will be stored in a separate secure cabinet. The digital recorder containing recorded participant interviews will be stored in a further separate secured filing cabinet in the same location. Audio recordings will be destroyed once each interview has been transcribed (estimated time: within one week). Transcripts generated from the study will be typed up and stored the NHS Fife secure IT network, which meets all relevant standards. This data will be fully anonymised and only consist of unique participant codes. Only anonymised data will be transferred for further analysis.

All data will be kept in accordance with the University of Edinburgh information governance procedures, and Good Clinical Practice Guidelines. To ensure confidentiality, participant identifiable information will be omitted from appearing on written transcriptions of interviews. Transcripts will

be anonymised and unique identification numbers will be assigned. The limits of confidentiality (i.e. if risk to self or other is identified the researcher may need to break confidentiality) will be explained to participants at the time they consent to taking part in the study. Any issues that may arise relating to data management confidentiality will be discussed with the academic and clinical supervisors of the study.

DISSEMINATION OF FINDINGS

Findings from this research will form part of the principal researcher's doctoral thesis in Clinical Psychology. It is also hoped that the study and findings will be submitted to peer reviewed scientific journals for publication. Outcomes will be shared with the CF teams who have taken part in the research with a view to broadening knowledge and considering clinical practice. Findings will also be distributed internally within the NHS (e.g. team meetings and presentation at the Fife Psychology Department conference). A summary of the findings will be written up and made available to the participants who have consented to being contacted with the research findings.

REFERENCES

- Berge, J. M., & Patterson, J. M. (2004). Cystic fibrosis and the family: A review and critique of the literature. *Families, Systems, & Health, 22*(1)(74).
- Brocki, J. M., & Wearden, A. J. (2006). A critical evaluation of the use of interpretative phenomenological analysis (IPA) in Health Psychology (Evaluating the use of IPA in Health Psychology). *Qualitative Research in Psychology. SAGE Benchmarks in Psychology Sage, 21*(31), 87–108. Retrieved from http://eprints.hud.ac.uk/id/eprint/10368/5/Brocki_and_Wearden_IPA.pdf
- Castellani, C., Southern, K. W., Brownlee, K., Roelse, J. D., Duff, A., Farrell, M., ... Elborn, S. (2009). European best practice guidelines for cystic fibrosis neonatal screening. *Journal of Cystic Fibrosis, 8*, 153–173. <http://doi.org/10.1016/j.jcf.2009.01.004>
- Chenail, R. J. (2011). Ten steps for conceptualizing and conducting qualitative research studies in a pragmatically curious manner. *The Qualitative Report, 16*(6), 1713–1730.
- Chow, M. Y. K., Morrow, A. M., Cooper Robbins, S. C., & Leask, J. (2013). Condition-specific quality of life questionnaires for caregivers of children with pediatric conditions: a systematic review. *Quality of Life Research, 22*(8), 2183–2200. <http://doi.org/10.1007/s11136-012-0343-z>
- Chudleigh, J., Buckingham, S., Dignan, J., O'Driscoll, S., Johnson, K., Rees, D., ... Metcalfe, A. (2016). Parents' Experiences of Receiving the Initial Positive Newborn Screening (NBS) Result for Cystic Fibrosis and Sickle Cell Disease. *Journal of Genetic Counseling, 25*(6), 1215–1226. <http://doi.org/10.1007/s10897-016-9959-4>
- Cousino, M. K., & Hazen, R. A. (2013). Parenting Stress Among Caregivers of Children With Chronic Illness: A Systematic Review. *Journal of Pediatric Psychology, 38*(8), 809–828. <http://doi.org/10.1093/jpepsy/jst049>

- Cystic Fibrosis Trust. (2018). What is cystic fibrosis?
<http://doi.org/https://www.cysticfibrosis.org.uk/what-is-cystic-fibrosis>
- Eddy, M. E., Carter, B. D., Kronenberger, W. G., Conradsen, S., Eid, N. S., Bourland, S. L., & Adams, G. (1998). Parent relationships and compliance in cystic fibrosis. *Journal of Pediatric Health Care : Official Publication of National Association of Pediatric Nurse Associates & Practitioners*, 12(4), 196–202. [http://doi.org/10.1016/S0891-5245\(98\)90046-3](http://doi.org/10.1016/S0891-5245(98)90046-3)
- Finlay, L. (2011). Phenomenology for Therapists Researching the Lived World Library of Congress Cataloging-in-Publication Data. Retrieved from www.wiley.com/wiley-blackwell.
- Fitzgerald, C., George, S., Somerville, R., Linnane, B., & Fitzpatrick, P. (2018). Caregiver burden of parents of young children with cystic fibrosis. *Journal of Cystic Fibrosis*, 17(1), 125–131. <http://doi.org/10.1016/J.JCF.2017.08.016>
- Glasscoe, C. A., & Quittner, A. L. (2008). Psychological interventions for people with cystic fibrosis and their families. In C. A. Glasscoe (Ed.), *Cochrane Database of Systematic Reviews*. Chichester, UK: John Wiley & Sons, Ltd. <http://doi.org/10.1002/14651858.CD003148.pub2>
- Havermans, T., Tack, J., Vertommen, A., Proesmans, M., & de Boeck, K. (2015). Breaking bad news, the diagnosis of cystic fibrosis in childhood. *Journal of Cystic Fibrosis : Official Journal of the European Cystic Fibrosis Society*, 14(4), 540–6. <http://doi.org/10.1016/j.jcf.2014.12.005>
- Hayeems, R. Z., Miller, F. A., Barg, C. J., Bombard, Y., Kerr, E., Tam, K., ... Guttman, A. (2016). Parent Experience With False-Positive Newborn Screening Results for Cystic Fibrosis. *Pediatrics*, 138(3).
- Helton, J. L., Harmon, R. J., Robinson, N., & Accurso, F. J. (1991). Parental attitudes toward newborn screening for cystic fibrosis. *Pediatric Pulmonology*, 11(S7), 23–28. <http://doi.org/10.1002/ppul.1950110706>
- Hodgkinson, R., & Lester, H. (2002). Stresses and coping strategies of mothers living with a child with cystic fibrosis: implications for nursing professionals. *Journal of Advanced Nursing*, 39(4), 377–383. <http://doi.org/10.1046/j.1365-2648.2002.02299.x>
- Holmes, A. M., & Deb, P. (2003). The Effect of Chronic Illness on the Psychological Health of Family Members. *The Journal of Mental Health Policy and Economics J Ment Health Policy Econ*, 6, 13–22. Retrieved from <https://pdfs.semanticscholar.org/1896/d37654641eb50ac569786470d20eeaba31b4.pdf>
- Howard, L. M., Piot, P., & Stein, A. (2014). No health without perinatal mental health. *The Lancet*, 384(9956), 1723–1724. [http://doi.org/10.1016/S0140-6736\(14\)62040-7](http://doi.org/10.1016/S0140-6736(14)62040-7)
- Jamieson, N., Fitzgerald, D., Singh-Grewal, D., Hanson, C. S., Craig, J. C., & Tong, A. (2014). Children's experiences of cystic fibrosis: a systematic review of qualitative studies. *Pediatrics*, 133(6), e1683-97. <http://doi.org/10.1542/peds.2014-0009>
- Jessup, M., Douglas, T., Priddis, L., Branch-Smith, C., & Shields, L. (2016). Parental Experience of Information and Education Processes Following Diagnosis of Their Infant With Cystic Fibrosis Via Newborn Screening. *Journal of Pediatric Nursing*, 31(3), e233–e241. <http://doi.org/10.1016/j.pedn.2015.11.010>
- National Health Service UK. (2018a). Cystic fibrosis. Retrieved March 17, 2018, from <https://www.nhs.uk/conditions/cystic-fibrosis/>

- National Health Service UK. (2018b). Newborn blood spot test. Retrieved March 17, 2018, from <https://www.nhs.uk/conditions/pregnancy-and-baby/newborn-blood-spot-test/>
- Parsons, E. P., & Bradley, D. M. (2003). Psychosocial issues in newborn screening for cystic fibrosis. *Paediatric Respiratory Reviews*, 4(4), 285–292. [http://doi.org/10.1016/S1526-0542\(03\)00086-1](http://doi.org/10.1016/S1526-0542(03)00086-1)
- Phares, V., Lopez, E., Fields, S., Kamboukos, D., & Duhig, A. M. (2005). Are Fathers Involved in Pediatric Psychology Research and Treatment? *Journal of Pediatric Psychology*, 30(8), 631–643. <http://doi.org/10.1093/jpepsy/jsi050>
- Quittner, A. L., Barker, D. H., Geller, D., Butt, S., & Gondor, M. (2007). 319 Effects of maternal depression on electronically monitored enzyme adherence and changes in weight for children with CF. *Journal of Cystic Fibrosis*, 6(S78).
- Quittner, A. L., Goldbeck, L., Abbott, J., Duff, A., Lambrecht, P., Solé, A., ... Barker, D. (2014). Prevalence of depression and anxiety in patients with cystic fibrosis and parent caregivers: results of The International Depression Epidemiological Study across nine countries. *Thorax*, 69(12), 1090–1097. <http://doi.org/10.1136/THORAXJNL-2014-205983>
- Rouf, K., White, L., & Evans, K. (2012). A qualitative investigation into the maternal experience of having a young child with severe food allergy. *Clinical Child Psychology and Psychiatry*, 17(171), 49–64. <http://doi.org/10.1177/1359104511415636>
- Schweitzer, R., Griffiths, M., & Yates, P. (2012). Parental experience of childhood cancer using Interpretative Phenomenological Analysis. *Psychology and Health*, 27(6), 704–720. <http://doi.org/10.1080/08870446.2011.622379>
- Scottish Government. (2015). Newborn Screening.
- Smith, B. A., Modi, A. C., Quittner, A. L., & Wood, B. L. (2010). Depressive symptoms in children with cystic fibrosis and parents and its effects on adherence to airway clearance. *Pediatric Pulmonology*, 45(8), 756–763. <http://doi.org/10.1002/ppul.21238>
- Smith, J. A. (2011). Evaluating the contribution of interpretative phenomenological analysis. *Health Psychology Review*, 5(1), 9–27. <http://doi.org/10.1080/17437199.2010.510659>
- Smith, J. A., Flowers, P., & Larkin, M. (2009). *Interpretative phenomenological analysis : theory, method, and research*. SAGE.
- Smyth, A., Bell, S. C., Bojcin, S., Bryon, M., Duff, A., Flume, P., ... Wolfe, S. (2014). European Cystic Fibrosis Society Standards of Care: Best Practice guidelines. *Journal of Cystic Fibrosis*, 13, S23–S42. <http://doi.org/10.1016/j.jcf.2014.03.010>
- Southern, K. W., Mérelle, M. M. E., Dankert-Roelse, J. E., & Nagelkerke, A. (2009). Newborn screening for cystic fibrosis. *Cochrane Database of Systematic Reviews*. <http://doi.org/10.1002/14651858.CD001402.pub2>
- The Butterfly Trust. (2018). Cystic Fibrosis: about the illness. Retrieved March 17, 2018, from <http://www.butterflytrust.org.uk/cystic-fibrosis/>
- Ulph, F., Cullinan, T., Qureshi, N., & Kai, J. (2015). Parents' responses to receiving sickle cell or cystic fibrosis carrier results for their child following newborn screening. *European Journal of Human Genetics*, 23(4), 459–465. <http://doi.org/10.1038/ejhg.2014.126>

Appendix N: Example of Transcribed Material and Themes

Example from one participant for superordinate theme: Cognitive and Emotional Experiences.

	Direct quotations	Summary of comments	Page
Cognitive and Emotional Experiences			
Vivid memories	"So I met his dad mmm just kind of when we were out socialising and stuff ... so really when we met, just like any young people ... family life was good ... life was good".	Looking back life good, like any other couple	1
	"And then the phone rang and that was the changer. And that was the hospital and it was the doctor to say that they got the test [child] had had when he was in the hospital."	Stands out as specific moment of change	2
	"So we went in for the appointment which was half past two. I can still remember that. And [the doctor] was right across from me, his dad was sitting next to me, my mum was next to me [gesturing] and CF nurse was in the room."	Clear specifics of times, where people were sitting	3
	"Because that was morning, [time] and we were to go in for [time]."	Clear memory of specific times	4
	"So definitely hit home quite quick that things were gonnae change quite dramatically."	Dramatic change hitting home	4
	"I can remember exactly what he was wearing – a blue babysuit and home knitted hat."	Recalling specifics of outfit	9
	"I guess all of sudden your world is turned upside down."	Everything changed in a moment	9
	"... cos when it was all getting says to me it's always that room I just remember... I can still sort of see what the room looks like and where everybody was sitting and how I was feeling at the time...I remember it being a very long afternoon."	Remembering what the room looked like and feelings she had	9
Psychological and emotional experiences	"It was quite an emotional time, it had nae properly sank in. I was quite shocked."	Emotional, shock	3
	"It was a shock."	Shock	4
	"Mmmm so it was pretty devastating at the time.... Definitely."	Definite devastation	4
	"It was a big impact on us all. We were all so shocked at the time."	Shock across the family	7

	"Like after I got over than initial shock and after I'd had the cry we got moved to the other room to get the sweat test and it maybe came more to life again."	Shock and crying	9
	"It was tough. It was really tough when you get home because you're trying to take it all in."	Tough situation, trying to take in info	9
	"I never knew what CF was so when I was told I was obviously shocked and googling it and what not."	Shock, unaware of CF	11
	"my mind was going a hundred miles an hour"	Mind racing	3
	"I was quite scared at what was to come."	Anxiety/scared	5
	"I only really noticed that when I had [other child], I was a lot more sort of laid back and less worried about things... I was just sort of more scared with [child]"	Worries highlighted with hindsight	5
	"I was quite scared at what was to come."	Anxiety/scared	5
	"It was tough. I thought I could do it all myself but it gets on top of you. I kind of stopped going out for a while. I mean just even for walks and things. I'd say I probably had a wee bit depression but I never went and tried to get anything for it or what not. I've just always kind of thought I'll get on with it and get through it. It was really hard, definitely."	?Isolation, low mood, depression	6
	"And you're just kind of sitting in a daze."	In a daze	9
Trying to manage uncertainty	"Sometimes you just over think it and it kind of beats you."	Overthinking/anxiety	8
	"because every CF kid/patient is different in how they keep, how things are going to be. It's hard to say. Like from one to the other who will keep better and who will nae. It's just what your body is like and stuff."	Variability between CF patients, can't predict	3
	"I knew nothing about genetics and I thought that maybe because of the birth he'd had maybe it detected something with the mucous and maybe that's just what's happened and it would be a misunderstanding."	Trying to figure out what it could be, looking for other explanations	3
	"But I think still at the back of mind I was nae convinced at the time"	Not convinced	3
	"I guess you just take things for granted and don't think it's going to happen to you."	Don't think it will happen to you, ?disbelief	7
	"I remember looking at him and thinking this is nae real, I'm gonna wake up and this has got to be a dream or something."	Disbelief/denial	9
	"Mmmm. I guess just wishing that it was nae real but unfortunately it was."	Wishing it was not real	9

Sense-making with hindsight	“Like when he was feeding bringing the milk back up and kind of shocking the mucous and fluids and stuff. We had to put him upside down and it was quite a worry because I kept thinking he was going to choke.”	Mucous, being unwell	1
	“Obviously there stuff about lungs and stuff but that was the bit that stuck in my head. And when I saw it said it was mucous in the lungs. I knew nothing about genetics and I thought that maybe because of the birth he’d had maybe it detected something with the mucous and maybe that’s just what’s happened and it would be a misunderstanding.”	Making link with symptoms and diagnosis	3
	“So I think he was six weeks old. He had nae in that time like before the diagnosis recognised anything bar the trauma from when he was born and all the stuff he had swallowed. His poo and stuff was more explosive but I just put that down to him being a new born baby and maybe the milk not being quite right. .”	Toileting issues	2

Appendix O: Excerpts from Reflective Journal

A reflective journal was kept during the data collection and analysis parts of this study. Excerpts from different phases are provided as examples here.

Excerpt 1. From interview phase, apprehension with beginning interviews and trying to find new role as researcher instead of clinician. Quite fixed on asking “right” questions at this point:

Completed first interview today and was feeling quite nervous. Had been checking over the interview questions before hand to try and be able to ask these questions without looking too much at notes. Noticed the pull to ask more “clinical” questions and took some conscious effort to step back from clinician role. Struck by how much this family is dealing with and how much information this mother was willing to share with me. Feels that I have been let into something very private, maybe especially because I am in their home? Noticed myself feeling a bit more on edge when the topic of conversation steered away from diagnosis but trying to sit with that and listen to what is important for mother. Speaking about lots of different times, not just diagnosis, I wonder if others will do the same. I hope there’s enough re diagnosis.

Excerpt 2. After transcribing an interview near the middle of the process. Some sense of frustration at the time it was taking but perhaps some hope that some commonalities were emerging.

Transcription is harder and taking longer than I anticipated. Finding it quite demanding to stay focused. Feel sense of responsibility to get exact wordings across

e.g. colloquialisms. Listening back, hearing more than I noticed at the time. Starting to feel there may be some overlaps here.

Excerpt 3: Supervision, review of emerging themes. Again, as someone new to IPA, apprehensive about doing it “correctly”.

Supervision to discuss emerging themes, was feeling worried as to whether I have done it right or not. Discussed this needing to do it “right”, Mark reassuring re approach. Reviewed quotes that may fit emerging themes and were in agreement. Feels like I can move on with the next step of paper now.

Excerpt 4: Following a feedback phone call with a participant. Content with interaction with parent and her feedback. Noticing her comment that it felt “therapeutic” to speak about diagnosis.

Feeling quite positive after speaking to X. She was agreeing as I was speaking and it felt like interpretations seemed fair and accurate to her. Very happy about this! Feels like an important step and have sense of relief. Nice to touch base again and get feedback of how she has been since interview. Said it was “therapeutic” to talk about it. Value in telling story in safe space perhaps?

Whole Thesis Portfolio References

- Ahmed, K. J., Ahmed, M., Jafri, H. S., Raashid, Y., & Ahmed, S. (2015). Pakistani mothers' and fathers' experiences and understandings of the diagnosis of Down syndrome for their child. *Journal of community genetics*, 6(1), 47-53. doi:<https://doi.org/10.1007/s12687-014-0200-6>
- Attar, M. A., Hernandez, M. E., Mullan, P. B., Tang, T. S., & Haftel, H. (2010). Pediatric residents' competency in communicating bad news and eliciting spiritual needs: Development and evaluation of a skills-based curriculum. *Journal of Neonatal-Perinatal Medicine*, 3(3), 177-185. doi:10.3233/NPM-2010-0113
- Ayers, S. L., & Kronenfeld, J. J. (2007). Chronic illness and health-seeking information on the Internet. *Health*, 11(3), 327-347. doi:<https://doi.org/10.1177/1363459307077547>
- Baile, W. F., Buckman, R., Lenzi, R., Glober, G., Beale, E. A., & Kudelka, A. P. (2000). SPIKES—a six-step protocol for delivering bad news: application to the patient with cancer. *The oncologist*, 5(4), 302-311. doi:10.1634/theoncologist.5-4-302
- Berge, J. M., & Patterson, J. M. (2004). Cystic fibrosis and the family: A review and critique of the literature. *Families, Systems, & Health*, 22(1), 74. doi:<https://doi.org/10.1037/1091-7527.22.1.74>
- Berkhof, M., van Rijssen, H. J., Schellart, A. J., Anema, J. R., & van der Beek, A. J. (2011). Effective training strategies for teaching communication skills to physicians: an overview of systematic reviews. *Patient education and counseling*, 84(2), 152-162. doi:<https://doi.org/10.1016/j.pec.2010.06.010>
- Besier, T., Born, A., Henrich, G., Hinz, A., Quittner, A. L., Goldbeck, L., & Group, T. S. (2011). Anxiety, depression, and life satisfaction in parents caring for children with cystic fibrosis. *Pediatric pulmonology*, 46(7), 672-682. doi:<https://doi.org/10.1002/ppul.21423>
- Biggerstaff, D., & Thompson, A. R. (2008). Interpretative phenomenological analysis (IPA): A qualitative methodology of choice in healthcare research. *Qualitative research in psychology*, 5(3), 214-224. doi:<https://doi.org/10.1080/14780880802314304>
- Bion, W. R. (1962). *Learning from Experience*. . London/New York: Karnac.

- Brown, R., Dunn, S., Byrnes, K., Morris, R., Heinrich, P., & Shaw, J. (2009). Doctors' stress responses and poor communication performance in simulated bad-news consultations. *Academic Medicine*, 84(11), 1595-1602.
doi:10.1097/ACM.0b013e3181baf537
- Brown, R., & Kulik, J. (1977). Flashbulb memories. *Cognition*, 5(1), 73-99.
doi:https://doi.org/10.1016/0010-0277(77)90018-X
- Buckman, R. (1984). Breaking bad news: why is it still so difficult? *British medical journal (Clinical research ed.)*, 288(6430), 1597.
doi:10.1136/bmj.288.6430.1597
- Cabizuca, M., Mendlowicz, M., Marques-Portella, C., Ragoni, C., Coutinho, E. S. F., de Souza, W., . . . Figueira, I. (2010). The invisible patients: posttraumatic stress disorder in parents of individuals with cystic fibrosis. *Revista de Psiquiatria Clínica*, 37(1), 6-17. Retrieved from
file:///C:/Users/blath/Downloads/17319-Article%20Text-20792-1-10-20120522%20(2).pdf
- Calhoun, A. W., Sutton, E. R., Barbee, A. P., McClure, B., Bohnert, C., Forest, R., . . . Fallat, M. E. (2017). Compassionate Options for Pediatric EMS (COPE): addressing communication skills. *Prehospital Emergency Care*, 21(3), 334-343. doi:https://doi.org/10.1080/10903127.2016.1263370
- Cannone, D., Atlas, M., Fornari, A., Barilla-LaBarca, M.-L., & Hoffman, M. (2019). Delivering Challenging News: An Illness-Trajectory Communication Curriculum for Multispecialty Oncology Residents and Fellows. *MedEdPORTAL: the journal of teaching and learning resources*, 15.
doi:10.15766/mep_2374-8265.10819
- Carpenter, K., Wittkowski, A., Hare, D. J., Medford, E., Rust, S., Jones, S. A., & Smith, D. M. (2018). Parenting a child with phenylketonuria (PKU): an interpretative phenomenological analysis (IPA) of the experience of parents. *Journal of genetic counseling*, 27(5), 1074-1086.
doi:https://doi.org/10.1007/s10897-018-0227-7
- Carter, N., Bryant-Lukosius, D., DiCenso, A., Blythe, J., & Neville, A. J. (2014). *The use of triangulation in qualitative research*. Paper presented at the Oncology nursing forum.
- Castellani, C., Southern, K. W., Brownlee, K., Roelse, J. D., Duff, A., Farrell, M., . . . Sermet-Gaudelus, I. (2009). European best practice guidelines for cystic

- fibrosis neonatal screening. *Journal of cystic fibrosis*, 8(3), 153-173.
doi:<http://doi.org/10.1016/j.jcf.2009.01.004>
- Chenail, R. J. (2011). Ten Steps for Conceptualizing and Conducting Qualitative Research Studies in a Pragmatically Curious Manner. *Qualitative Report*, 16(6), 1713-1730. doi:<https://files.eric.ed.gov/fulltext/EJ956240.pdf>
- Choudhary, A., & Gupta, V. (2015). Teaching communications skills to medical students: Introducing the fine art of medical practice. *International Journal of Applied and Basic Medical Research*, 5(Suppl 1), S41. doi:10.4103/2229-516X.162273
- Chow, M. Y. K., Morrow, A. M., Robbins, S. C. C., & Leask, J. (2013). Condition-specific quality of life questionnaires for caregivers of children with pediatric conditions: A systematic review. *Quality of Life Research*, 22(8), 2183-2200. doi:<https://doi.org/10.1007/s11136-012-0343-z>
- Chudleigh, J., Buckingham, S., Dignan, J., O'Driscoll, S., Johnson, K., Rees, D., . . . Metcalfe, A. (2016). Parents' experiences of receiving the initial positive newborn screening (NBS) result for cystic fibrosis and sickle cell disease. *Journal of genetic counseling*, 25(6), 1215-1226. doi:10.1258/jrsm.2008.s18006.
- Clancy, M. (2013). Is reflexivity the key to minimising problems of interpretation in phenomenological research? *Nurse researcher*, 20(6). doi:10.7748/nr2013.07.20.6.12.e1209
- Collins, J. L., La Pean, A., O'Tool, F., Eskra, K. L., Roedl, S. J., Tluczek, A., & Farrell, M. H. (2013). Factors that influence parents' experiences with results disclosure after newborn screening identifies genetic carrier status for cystic fibrosis or sickle cell hemoglobinopathy. *Patient education and counseling*, 90(3), 378-385. doi:10.1016/j.pec.2011.12.007
- Coughlin, M. B., & Sethares, K. A. (2017). Chronic sorrow in parents of children with a chronic illness or disability: An integrative literature review. *Journal of pediatric nursing*, 37, 108-116. doi:<https://doi.org/10.1016/j.pedn.2017.06.011>
- Cousino, M. K., & Hazen, R. A. (2013). Parenting stress among caregivers of children with chronic illness: a systematic review. *Journal of pediatric psychology*, 38(8), 809-828. doi:<http://doi.org/10.1093/jpepsy/jst049>
- Cystic Fibrosis Trust. (2020). What is cystic fibrosis? Retrieved from <https://www.cysticfibrosis.org.uk/what-is-cystic-fibrosis>

- Davies, B., & Connaughty, S. (2002). Pediatric end-of-life care: Lessons learned from parents. *JONA: The Journal of Nursing Administration*, 32(1), 5-6.
- Deave, T., & Johnson, D. (2008). The transition to parenthood: what does it mean for fathers? *Journal of advanced nursing*, 63(6), 626-633.
doi:<https://doi.org/10.1111/j.1365-2648.2008.04748.x>
- Deeks, J. J., Dinnes, J., D'amico, R., Sowden, A. J., Sakarovitch, C., Song, F., . . . Altman, D. (2003). Evaluating non-randomised intervention studies. *Health technology assessment*, 7(27), 1-179. doi:<https://doi.org/10.3310/hta7270>
- Dillard, J. P., Shen, L., Robinson, J. D., & Farrell, P. M. (2010). Parental information seeking following a positive newborn screening for cystic fibrosis. *Journal of health communication*, 15(8), 880-894.
doi:<https://doi.org/10.1080/10810730.2010.522226>
- Dosanjh, S., Barnes, J., & Bhandari, M. (2001). Barriers to breaking bad news among medical and surgical residents. *Medical education*, 35(3), 197-205.
doi:<https://doi.org/10.1111/j.1365-2923.2001.00766.x>
- Doss, B. D., Rhoades, G. K., Stanley, S. M., & Markman, H. J. (2009). The effect of the transition to parenthood on relationship quality: an 8-year prospective study. *Journal of personality and social psychology*, 96(3), 601.
doi:<https://doi.org/10.1037/a0013969>
- Douglas, H. (2007). *Containment and reciprocity: Integrating psychoanalytic theory and child development research for work with children*: Routledge.
- Downs, S. H., & Black, N. (1998). The feasibility of creating a checklist for the assessment of the methodological quality both of randomised and non-randomised studies of health care interventions. *Journal of Epidemiology & Community Health*, 52(6), 377-384. doi:<http://dx.doi.org/10.1136/jech.52.6.377>
- Dubé, C. E., LaMonica, A., Boyle, W., Fuller, B., & Burkholder, G. J. (2003). Self-assessment of communication skills preparedness: adult versus pediatric skills. *Ambulatory pediatrics*, 3(3), 137-141. doi:[https://doi.org/10.1367/1539-4409\(2003\)003<0137:SOCSPA>2.0.CO;2](https://doi.org/10.1367/1539-4409(2003)003<0137:SOCSPA>2.0.CO;2)
- Fallowfield, L., & Jenkins, V. (2004). Communicating sad, bad, and difficult news in medicine. *The Lancet*, 363(9405), 312-319. doi:[https://doi.org/10.1016/S0140-6736\(03\)15392-5](https://doi.org/10.1016/S0140-6736(03)15392-5)
- Finlay, L. (2011). *Phenomenology for therapists: Researching the lived world*: John Wiley & Sons.

- Fitzgerald, C., George, S., Somerville, R., Linnane, B., & Fitzpatrick, P. (2018). Caregiver burden of parents of young children with cystic fibrosis. *Journal of cystic fibrosis*, 17(1), 125-131. doi:<http://doi.org/10.1016/J.JCF.2017.08.016>
- Forrest, L. E., Curnow, L., Delatycki, M. B., Skene, L., & Aitken, M. (2008). Health first, genetics second: exploring families' experiences of communicating genetic information. *European Journal of Human Genetics*, 16(11), 1329-1335. doi:<https://doi.org/10.1038/ejhg.2008.104>
- General Medical Council. (2013). *Good Medical Practice*. General Medical Council Retrieved from https://www.gmc-uk.org/-/media/documents/good-medical-practice---english-20200128_pdf-51527435.pdf?la=en&hash=DA1263358CCA88F298785FE2BD7610EB4EE9A530
- Greenberg, L. W., Ochsenschlager, D., O'Donnell, R., Mastruserio, J., & Cohen, G. J. (1999). Communicating bad news: a pediatric department's evaluation of a simulated intervention. *Pediatrics*, 103(6), 1210-1217. doi:<https://doi.org/10.1542/peds.103.6.1210>
- Havermans, T., Tack, J., Vertommen, A., Proesmans, M., & de Boeck, K. (2015). Breaking bad news, the diagnosis of cystic fibrosis in childhood. *Journal of cystic fibrosis*, 14(4), 540-546. doi:<http://doi.org/10.1016/j.jcf.2014.12.005>
- Hayeems, R. Z., Miller, F. A., Barg, C. J., Bombard, Y., Kerr, E., Tam, K., . . . Davies, C. (2016). Parent experience with false-positive newborn screening results for cystic fibrosis. *Pediatrics*, 138(3), e20161052. doi:DOI: <https://doi.org/10.1542/peds.2016-1052>
- Higgins, J. P., & Green, S. (2011). *Cochrane handbook for systematic reviews of interventions* (Vol. 4): John Wiley & Sons.
- Hodgkinson, R., & Lester, H. (2002). Stresses and coping strategies of mothers living with a child with cystic fibrosis: implications for nursing professionals. *Journal of advanced nursing*, 39(4), 377-383. doi:<http://doi.org/10.1046/j.1365-2648.2002.02299.x>
- Holmes, A. M., & Deb, P. (2003). The effect of chronic illness on the psychological health of family members. *Journal of Mental Health Policy and Economics*, 6(1), 13-22. Retrieved from <https://pdfs.semanticscholar.org/1896/d37654641eb50ac569786470d20eeaba31b4.pdf>

- Howard, L. M., Piot, P., & Stein, A. (2014). No health without perinatal mental health. *The Lancet*. doi:[http://doi.org/10.1016/S0140-6736\(14\)62040-7](http://doi.org/10.1016/S0140-6736(14)62040-7)
- Hulsman, R. L., Pranger, S., Koot, S., Fabriek, M., Karemaker, J. M., & Smets, E. M. (2010). How stressful is doctor–patient communication? Physiological and psychological stress of medical students in simulated history taking and bad-news consultations. *International Journal of Psychophysiology*, 77(1), 26-34. doi:<https://doi.org/10.1016/j.ijpsycho.2010.04.001>
- Imo, U. O. (2017). Burnout and psychiatric morbidity among doctors in the UK: a systematic literature review of prevalence and associated factors. *BJPsych bulletin*, 41(4), 197-204. doi:<https://doi.org/10.1192/pb.bp.116.054247>
- Irwin, S., & Elley, S. (2013). Parents' hopes and expectations for their children's future occupations. *The Sociological Review*, 61(1), 111-130. doi:<https://doi.org/10.1111/j.1467-954X.2012.02139.x>
- Iversen, A. E., Esbjørn, B. H., Christensen, E., & Hansen, N. S. (2012). Exploring the impact of involving fathers in the treatment of their children: A study using Interpretative Phenomenological Analysis (IPA). *Qualitative Studies*, 3(1), 22-39. doi:<https://doi.org/10.7146/qs.v3i1.6271>
- James, L., & Johnson, B. (1997). The needs of parents of pediatric oncology patients during the palliative care phase. *Journal of Pediatric Oncology Nursing*, 14(2), 83-95. doi:[https://doi.org/10.1016/S1043-4542\(97\)90006-0](https://doi.org/10.1016/S1043-4542(97)90006-0)
- Jedlicka-Köhler, I., Götz, M., & Eichler, I. (1996). Parents' recollection of the initial communication of the diagnosis of cystic fibrosis. *Pediatrics*, 97(2), 204-209.
- Jessup, M., Douglas, T., Priddis, L., Branch-Smith, C., & Shields, L. (2016). Parental experience of information and education processes following diagnosis of their infant with cystic fibrosis via newborn screening. *Journal of pediatric nursing*, 31(3), e233-e241. doi:<https://doi.org/10.1016/j.pedn.2015.11.010>
- Jessup, M., Smyth, W., Abernethy, G., Shields, L., Douglas, T., & AREST-CF. (2018). Family-centred care for families living with cystic fibrosis in a rural setting: A qualitative study. *Journal of clinical nursing*, 27(3-4), e590-e599. doi:<https://doi.org/10.1111/jocn.14105>
- Johnson, J., & Panagioti, M. (2018). Interventions to Improve the Breaking of Bad or Difficult News by Physicians, Medical Students, and Interns/Residents: A Systematic Review and Meta-Analysis. *Academic Medicine*, 93(9), 1400-1412. doi:10.1097/Acm.0000000000002308

- King, A., & Hoppe, R. B. (2013). "Best practice" for patient-centered communication: a narrative review. *Journal of graduate medical education*, 5(3), 385-393. doi:<https://doi.org/10.4300/JGME-D-13-00072.1>
- Lefebvre, C., Glanville, J., Briscoe, S., Littlewood, A., Marshall, C., Metzendorf, M.-I., . . . Wieland, L. (2019). Chapter 4: Searching for and selecting studies. In T. J. Higgins JPT, Chandler J, Cumpston M, Li T, Page MJ, Welch VA (Ed.), *Cochrane Handbook for Systematic Reviews of Interventions version 6.0 (updated July 2019)*. Cochrane, 2019.
- Levetown, M. (2008). Communicating with children and families: from everyday interactions to skill in conveying distressing information. *Pediatrics*, 121(5), e1441-e1460. doi:<https://doi.org/10.1542/peds.2008-0565>
- Mack, J. W., Hilden, J. M., Watterson, J., Moore, C., Turner, B., Grier, H. E., . . . Wolfe, J. (2005). Parent and physician perspectives on quality of care at the end of life in children with cancer. *J Clin Oncol*, 23(36), 9155-9161. doi:10.1200/JCO.2005.04.010
- Meyer, E. C., Ritholz, M. D., Burns, J. P., & Truog, R. D. (2006). Improving the quality of end-of-life care in the pediatric intensive care unit: parents' priorities and recommendations. *Pediatrics*, 117(3), 649-657. doi:<https://doi.org/10.1542/peds.2005-0144>
- Moher, D., Liberati, A., Tetzlaff, J., & Altman, D. G. (2009). Preferred reporting items for systematic reviews and meta-analyses: the PRISMA statement. *Annals of internal medicine*, 151(4), 264-269. doi:10.7326/0003-4819-151-4-200908180-00135
- Monden, K. R., Gentry, L., & Cox, T. R. (2016). Delivering bad news to patients. *Baylor University Medical Center Proceedings*, 29(1), 101-102. doi:<https://doi.org/10.1080/08998280.2016.11929380>
- Mowder, B. A. (2005). Parent development theory: Understanding parents, parenting perceptions and parenting behaviors. *Journal of Early Childhood and Infant Psychology*, 1, 45-65.
- Muñoz Sastre, M., Sorum, P., & Mullet, E. (2014). Lay people's and health professionals' views about breaking bad news to children. *Child: care, health and development*, 40(1), 106-114. doi:<https://doi.org/10.1111/j.1365-2214.2012.01420.x>

- Narayanan, V., Bista, B., & Koshy, C. (2010). 'BREAKS' protocol for breaking bad news. *Indian journal of palliative care*, 16(2), 61. doi:10.4103/0973-1075.68401
- National Health Service. (2018). Cystic fibrosis. Retrieved from <https://www.nhs.uk/conditions/cystic-fibrosis/>
- National Institute for Health and Clinical Excellence. (2012). *Patient Experience in Adult NHS Services: Improving the Experience of Care for People Using Adult NHS Services (CG138)*. London, UK: National Institute for Health and Care Excellence Retrieved from <https://www.ncbi.nlm.nih.gov/books/NBK115230/>
- Nellis, M. E., Howell, J. D., Ching, K., & Bylund, C. (2017). The use of simulation to improve resident communication and personal experience at end-of-life care. *Journal of pediatric intensive care*, 6(02), 091-097. doi:10.1055/s-0036-1584684
- Northington, L. (2000). Chronic sorrow in caregivers of school age children with sickle cell disease: A grounded theory approach. *Issues in Comprehensive Pediatric Nursing*, 23(3), 141-154.
doi:<https://doi.org/10.1080/01460860050174693>
- Nunn, K. (2019). Delivering bad news. *Journal of paediatrics and child health*, 55(6), 617-620. doi:<https://doi.org/10.1111/jpc.14443>
- Ødegård, W. (2005). Chronic illness as a challenge to the attachment process. *Clinical child psychology and psychiatry*, 10(1), 13-22.
doi:<https://doi.org/10.1177/1359104505048787>
- Parsons, E. P., & Bradley, D. M. (2003). Psychosocial issues in newborn screening for cystic fibrosis. *Paediatric respiratory reviews*, 4(4), 285-292.
doi:[http://doi.org/10.1016/S1526-0542\(03\)00086-1](http://doi.org/10.1016/S1526-0542(03)00086-1)
- Patton, M. Q. (1999). Enhancing the quality and credibility of qualitative analysis. *Health services research*, 34(5 Pt 2), 1189.
- Ptacek, J., & Eberhardt, T. L. (1996). Breaking bad news: a review of the literature. *Jama*, 276(6), 496-502. doi:doi:10.1001/jama.1996.03540060072041
- Ptacek, J., & McIntosh, E. G. (2009). Physician challenges in communicating bad news. *Journal of behavioral medicine*, 32(4), 380-387.
doi:<https://doi.org/10.1007/s10865-009-9213-8>
- Quittner, A. L., Goldbeck, L., Abbott, J., Duff, A., Lambrecht, P., Solé, A., . . . Catastini, P. (2014). Prevalence of depression and anxiety in patients with

- cystic fibrosis and parent caregivers: results of The International Depression Epidemiological Study across nine countries. *Thorax*, 69(12), 1090-1097. doi:<http://dx.doi.org/10.1136/thoraxjnl-2014-205983>
- Rabow, M. W., & Mcphee, S. J. (1999). Beyond breaking bad news: how to help patients who suffer. *Western Journal of Medicine*, 171(4), 260-263.
- Ratcliffe, E., Pickering, S., McLean, S., & Lewis, J. (2014). Is there a relationship between subacromial impingement syndrome and scapular orientation? A systematic review. *Br J Sports Med*, 48(16), 1251-1256. doi:10.1136/bjsports-2013-092389corr1
- Redshaw, M., & Martin, C. (2014). The couple relationship before and during transition to parenthood. doi:<https://doi.org/10.1080/02646838.2014.896146>
- Reed, S., Kassis, K., Nagel, R., Verbeck, N., Mahan, J. D., & Shell, R. (2015). Breaking bad news is a teachable skill in pediatric residents: A feasibility study of an educational intervention. *Patient education and counseling*, 98(6), 748-752. doi:<https://doi.org/10.1016/j.pec.2015.02.015>
- Rider, E. A., Volkan, K., & Hafler, J. P. (2008). Pediatric residents' perceptions of communication competencies: Implications for teaching. *Medical teacher*, 30(7), e208-e217. doi:<https://doi.org/10.1080/01421590802208842>
- Rimé, B. (2007). The social sharing of emotion as an interface between individual and collective processes in the construction of emotional climates. *Journal of social issues*, 63(2), 307-322. doi:<https://doi.org/10.1111/j.1540-4560.2007.00510.x>
- Salm, N., Yetter, E., & Tluczek, A. (2012). Informing parents about positive newborn screen results: parents' recommendations. *Journal of Child Health Care*, 16(4), 367-381. doi:<https://doi.org/10.1177/1367493512443906>
- Sawicki, G. S., Heller, K. S., Demars, N., & Robinson, W. M. (2015). Motivating adherence among adolescents with cystic fibrosis: youth and parent perspectives. *Pediatric pulmonology*, 50(2), 127-136. doi:<https://doi.org/10.1002/ppul.23017>
- Scottish Government. (2019). Health Secretary welcomes cystic fibrosis drug pricing agreement. Retrieved from <https://www.gov.scot/news/health-secretary-welcomes-cystic-fibrosis-drug-pricing-agreement/>

- Shaw, J., Brown, R., Heinrich, P., & Dunn, S. (2013). Doctors' experience of stress during simulated bad news consultations. *Patient education and counseling*, 93(2), 203-208. doi:<https://doi.org/10.1016/j.pec.2013.06.009>
- Shaw, W. S., Zaia, A., Pransky, G., Winters, T., & Patterson, W. B. (2005). Perceptions of provider communication and patient satisfaction for treatment of acute low back pain. *Journal of Occupational and Environmental Medicine*, 47(10), 1036-1043. doi:10.1097/01.jom.0000172863.26222.14
- Silva, D. H. (2008). A competency-based communication skills workshop series for pediatric residents. *B LETÍN*, 8.
- Smith, J., Flowers, P., & Larkin, M. (2009). Planning an IPA research study. *Interpretative phenomenological analysis: theory, method and research*. London: SAGE Publications, 40-55.
- Smith, J. A. (2011). Evaluating the contribution of interpretative phenomenological analysis: A reply to the commentaries and further development of criteria. *Health psychology review*, 5(1), 55-61. doi:<http://doi.org/10.1080/17437199.2010.510659>
- Smith, J. A., Jarman, M., & Osborn, M. (1999). Doing interpretative phenomenological analysis. *Qualitative health psychology: Theories and methods*, 218-240.
- Smyth, A. R., Bell, S. C., Bojcin, S., Bryon, M., Duff, A., Flume, P., . . . Schwarzenberg, S. J. (2014). European cystic fibrosis society standards of care: best practice guidelines. *Journal of cystic fibrosis*, 13, S23-S42. doi:<https://doi.org/10.1016/j.jcf.2014.03.010>
- Southern, K. W., Merelle, M. M., Dankert-Roelse, J. E., & Nagelkerke, A. (2009). Newborn screening for cystic fibrosis. *Cochrane Database of Systematic Reviews*(1). doi:<http://doi.org/10.1002/14651858.CD001402.pub2>
- Stewart, M., Meredith, L., Brown, J. B., & Galajda, J. (2000). The influence of older patient-physician communication on health and health-related outcomes. *Clinics in geriatric medicine*, 16(1), 25-36, vii-viii. doi:10.1016/s0749-0690(05)70005-7
- Stewart, R., Hargreaves, K., & Oliver, S. (2004). Telling parents about the heel-prick test: consultation results and resource development. London: UK Newborn Screening Programme Centre. Retrieved from https://www.researchgate.net/profile/Ruth_Stewart/publication/42385256_Telli

ng_parents_about_the_heel-
prick_test_consultation_results_and_resources_development/links/00b7d513f
633ac8f57000000.pdf

- Street Jr, R. L., Makoul, G., Arora, N. K., & Epstein, R. M. (2009). How does communication heal? Pathways linking clinician–patient communication to health outcomes. *Patient education and counseling*, 74(3), 295-301. doi:<https://doi.org/10.1016/j.pec.2008.11.015>
- Street, R. L. (1991). Physicians' communication and parents' evaluations of pediatric consultations. *Medical care*, 29(11), 1146-1152.
- Tobler, K., Grant, E., & Marczyński, C. (2014). Evaluation of the impact of a simulation-enhanced breaking bad news workshop in pediatrics. *Simulation in Healthcare*, 9(4), 213-219. doi:10.1097/SIH.0000000000000031.
- Tools, N. C. C. f. M. a. (2008). Quality checklist for health care intervention studies.
- Tuffour, I. (2017). A critical overview of interpretative phenomenological analysis: a contemporary qualitative research approach. *Journal of Healthcare Communications*, 2(4), 52. doi:DOI: 10.4172/2472-1654.100093
- Tuohy, D., Cooney, A., Dowling, M., Murphy, K., & Sixsmith, J. (2013). An overview of interpretive phenomenology as a research methodology. *Nurse researcher*, 20(6). doi:10.7748/nr2013.07.20.6.17.e315
- Ulph, F., Cullinan, T., Qureshi, N., & Kai, J. (2015). Parents' responses to receiving sickle cell or cystic fibrosis carrier results for their child following newborn screening. *European Journal of Human Genetics*, 23(4), 459-465. doi:<http://doi.org/10.1038/ejhg.2014.126>
- Vaidya, V. U., Greenberg, L. W., Patel, K. M., Strauss, L. H., & Pollack, M. M. (1999). Teaching physicians how to break bad news: a 1-day workshop using standardized parents. *Archives of pediatrics & adolescent medicine*, 153(4), 419-422. doi:10.1001/archpedi.153.4.419
- Wanzer, M. B., Booth-Butterfield, M., & Gruber, K. (2004). Perceptions of health care providers' communication: relationships between patient-centered communication and satisfaction. *Health communication*, 16(3), 363-384. doi:https://doi.org/10.1207/S15327027HC1603_6
- Wennick, A., & Hallström, I. (2006). Swedish families' lived experience when a child is first diagnosed as having insulin-dependent diabetes mellitus: an ongoing

- learning process. *Journal of Family Nursing*, 12(4), 368-389.
doi:<https://doi.org/10.1177/1074840706296724>
- Wilson, B. (2020, December 1). Practical Meta-Analysis Effect Size Calculator.
<https://campbellcollaboration.org/escalc/html/EffectSizeCalculator-SMD3.php>
- Wong, M., & Heriot, S. (2008). Parents of children with cystic fibrosis: how they hope, cope and despair. *Child: care, health and development*, 34(3), 344-354.
doi: <https://doi.org/10.1111/j.1365-2214.2007.00804.x>
- Yuan, Y. Y., Scott, S., Van Horn, N., Oke, O., & Okada, P. (2019). Objective evaluation of a simulation course for residents in the pediatric emergency medicine department: breaking bad news. *Cureus*, 11(1).
doi:[10.7759/cureus.3903](https://doi.org/10.7759/cureus.3903)